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THE RENAL FASCIA

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THE descriptions of the arrangement of the renal fascia given in text-books of anatomy and surgical anatomy are more or less inaccurate, and apparently they are based mainly on the work of Zuckerkandl (1883), Sappey (1889), and Gerota (1895). The anterior and posterior layers of the fascia are said to be fused above and laterally, but to remain separate medially and below, and the anterior layers of the two sides are described as being directly continuous anterior to the great vessels. In effect this means that the two perinephric spaces are also continuous in front of the vertebral column, a conception illustrated in Gerota's diagram of the arrangement as seen in transverse section and repeated in many other diagrams in various articles and books. If this arrangement did, in fact, exist, any collection of blood, pus, or extravasated urine in one perinephric space could spread to the opposite side, yet every surgeon knows that such an occurrence is a clinical rarity.

Knight (1893) stated that the two layers of renal fascia united on the medial side around the renal vessels, and Poirier (1904) and Gosset (1907) asserted that the perinephric space of one side was not continuous with the other, pointing out that neither layer of renal fascia was directly continuous with its opposite number across the midline—unless, Gosset added, the kidneys were fused as in horseshoe kidney. They showed that the anterior layer blended with the connective tissue around the great vessels, in the root of the mesentery and around the pancreas, and that the posterior layer was fixed to the vertebræ anterior to the attachment of psoas major. They stated that inferiorly the two layers gradually became lost in the fascia of the iliac fossa. Southam (1923) provided further evidence in support of Poirier's and Gosset's views, and Mitchell (1939) and Martin (1942) recorded other investigations and experiments which all confirmed in the main the descriptions of the French writers; but the views of the earlier workers still prevail. Many more specimens have now been examined and additional information has been obtained about the exact disposition of the renal fascia and its connexions, and about the spread of fluids from the perinephric and paranephric spaces.

The true disposition of the fascia may be determined by routine anatomical procedures, and it is shown dramatically by radiological methods. A fine barium emulsion is injected slowly through a needle inserted into the perinephric space and the process is watched on the radiographic screen. Plates may be exposed at regular intervals or whenever an interesting appearance is detected, and they provide a permanent record. Subsequently the specimens are dissected, or sectioned transversely or vertically, to confirm the X-ray findings. Owing to the shortage of subjects all the experiments recorded here were performed on infants, but

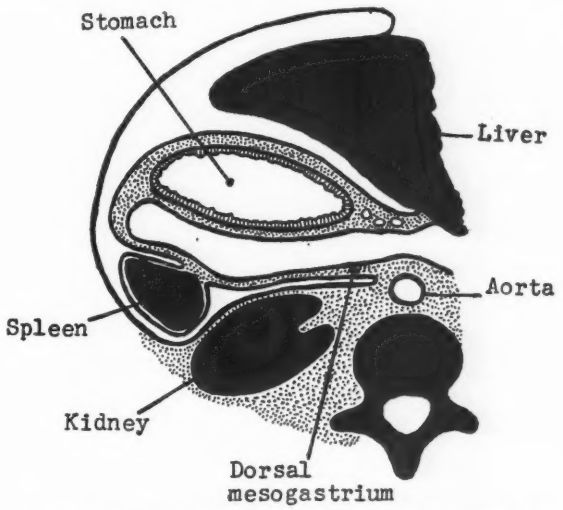


FIG. 329.—During development the dorsal mesogastrium becomes pressed against the posterior abdominal wall. Ultimately the opposed peritoneal layers fuse and their remains lie between the definitive layer of the peritoneum in this region and the structures behind it.

examination of nine adult dissecting-room subjects revealed no fundamental differences in the arrangement. Seventy-six radiological experiments have now been performed, but 12 of these were failures owing to the needle being inserted too far or not far enough, leaving 64 valid results for analysis.

Most writers are agreed on certain points. The renal fascia consists of anterior and posterior layers formed by a splitting of the retroperitoneal subserous tissue, and the posterior layer is more definite than the anterior as the latter is more or less firmly attached to the overlying peritoneum where they are in contact; where the kidneys are in relationship with adjacent viscera such as the colon, duodenum, and pancreas, the anterior layers fuse with the intervening connective tissue. The left anterior layer is rather thicker than the right because in the course of development the dorsal mesogastrium and left

primitive mesocolon become pressed against the posterior abdominal wall and ultimately the opposed peritoneal layers become obliterated and the remains

Both layers of renal fascia are separated from the true renal capsule by a variable amount of perinephric fat, and there is another variable deposit

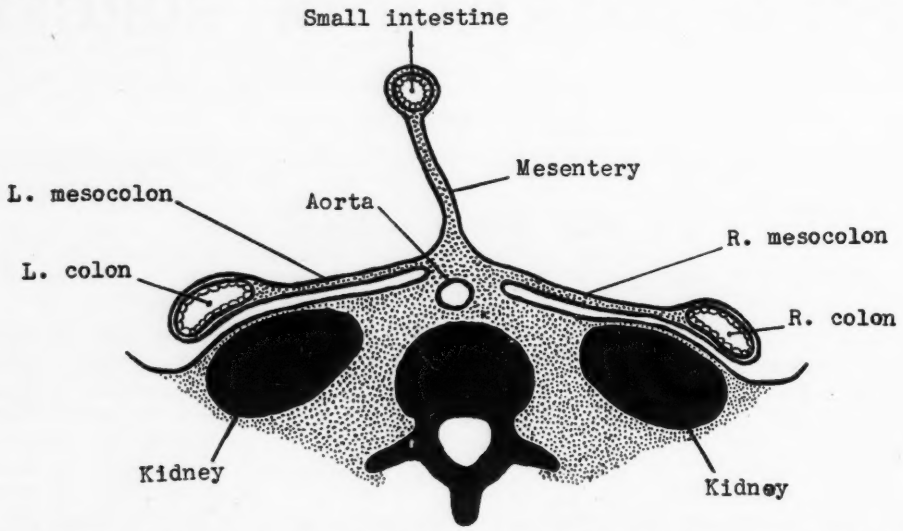
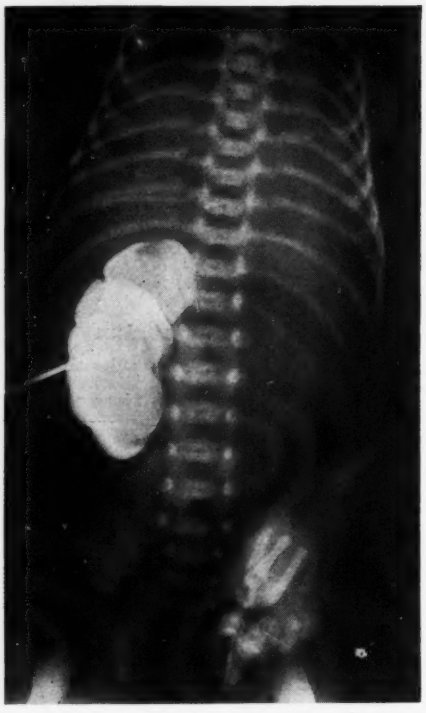
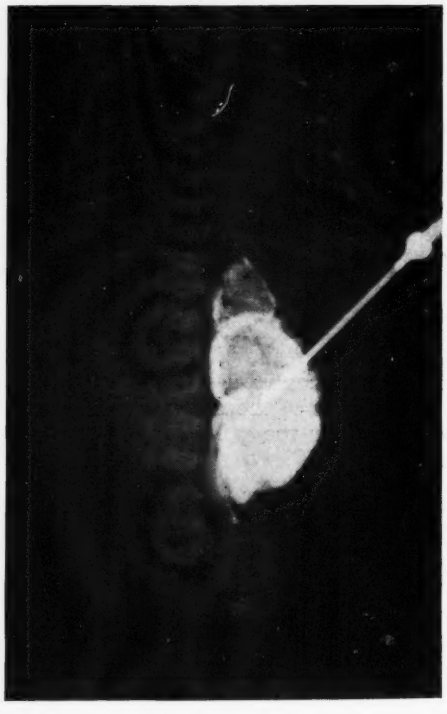


FIG. 330.—Originally both right and left colons possess a mesocolon, but during development these disappear as the adjacent layers of peritoneum fuse together.

lie partly between the definitive layer of peritoneum and the left kidney (*Figs.* 329, 330). On the right side the primitive mesocolon behaves in a similar fashion, but the colon is lower than on the left side and so a smaller part of its mesocolon lies in relationship to the right kidney, while there are no remnants of the dorsal mesogastrium in relationship to this kidney.

of fat behind the posterior layer of fascia in the paranephric region postero-inferior to the kidney. These fatty deposits are better developed in adults.





FIGS. 331, 332.—Note the typical reniform shape of the radiographic shadow and the coincident filling of the suprarenal space. Injection pressure 5–6 mm. Hg.

There is general agreement about the above points, and about the fact that the anterior and posterior layers are fused above and laterally, forming a loose common investment for the suprarenal and kidney. There are differences of opinion,

however, about the arrangement medially and inferiorly, the majority of writers expressing the view that the two layers remain separate in these situations. But there is good evidence that they are also united in these areas in a manner to be described hereafter, and this can be demonstrated graphically by the injection experiments.

FIG. 333.—A vertical section showing the barium emulsion enclosed within the fascia around the kidney and suprarenal. The spread above and below is limited by fusion of the layers of renal fascia in these situations. The small trickle of barium in the retroperitoneal tissues below the kidney escaped in the vicinity of the ureter.

When the barium emulsion is injected the radiographic shadow is soon observed to assume a typical reniform shape, and this alone is sufficient to show that the layers of fascia are united along the entire renal margin and not only above and laterally (*Figs.* 331, 332). At an early stage, and while the pressure is still low (5–6 mm. Hg) the space surrounding the suprarenal gland also becomes filled, showing that normally there is no complete fascial layer between the two organs (*Figs.* 331, 332). Sappey (1889), Knight (1893), von Bergmann (1904), Billington (1910), Vecchi (1910), Stromberg (1910), Tobin (1944), and others state that there is a thin layer of fascia between them, but Gosset (1907) denied this, and the most I have seen on

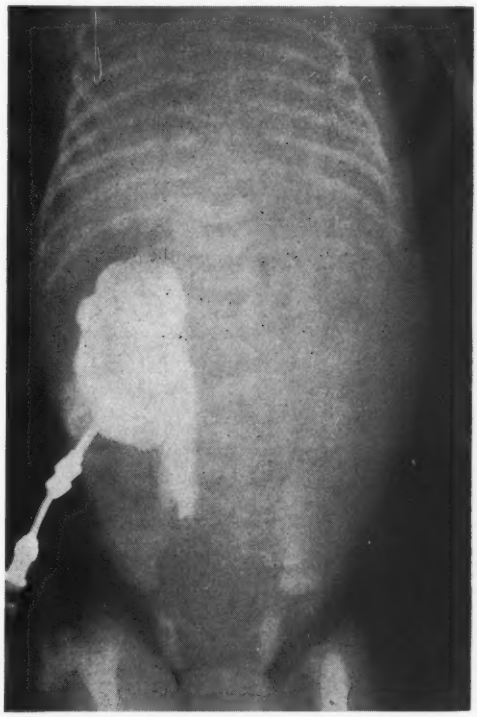


FIG. 334.—The barium has commenced to escape from the perinephric space in the vicinity of the ureter. Injection pressure 7 mm. Hg.

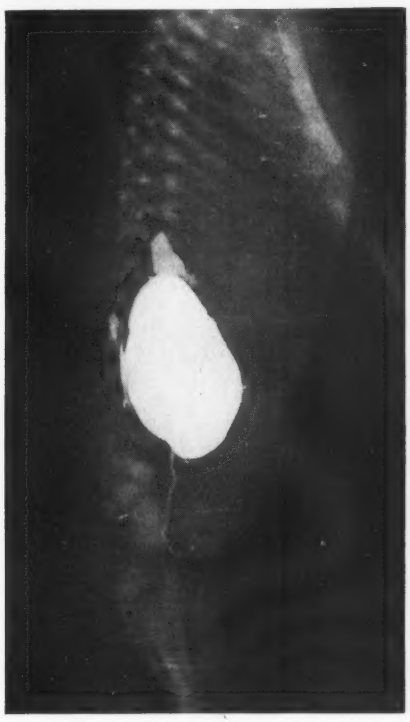


FIG. 335.—A lateral view showing barium escaping from the perinephric space around the ureter. The slight irregularity in the shape of the reniform shadow posteriorly is due to escape of barium along the needle track. Injection pressure 7 mm. Hg.

dissection is a thin perforate septum, except in two adult specimens in which an apparently complete layer existed. Speculation has been aroused by the fact that in cases of movable kidney the suprarenal retains its normal position. Probably this is due to the fact that it is much lighter and so less

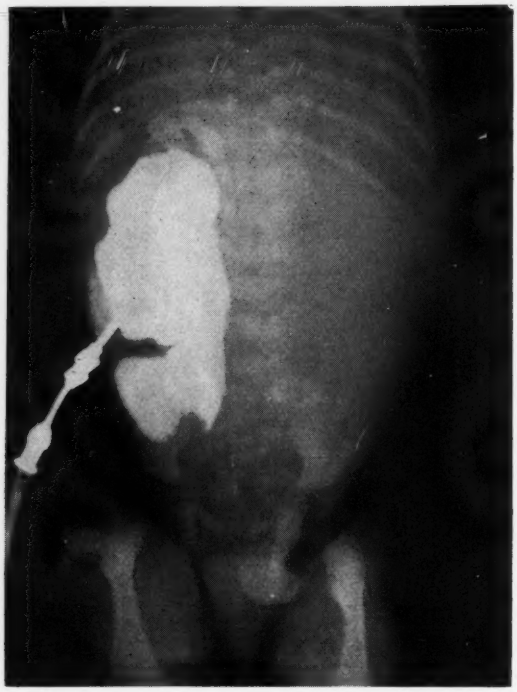


FIG. 336.—Note the clear interval between the barium in the perinephric space and the effused barium in the retroperitoneal tissues below the kidney. Injection pressure 9 mm. Hg.

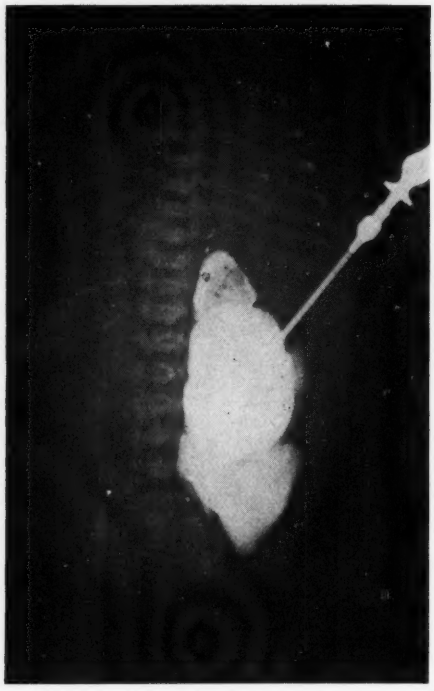


FIG. 337.—Most of the barium escaping from the perinephric space has done so in the vicinity of the ureter, but a small amount has escaped laterally, showing that the fusion between the layers of renal fascia on the lateral side is not always as firm as is commonly stated. At the time the radiograph was taken the injection pressure was 10 mm. Hg.

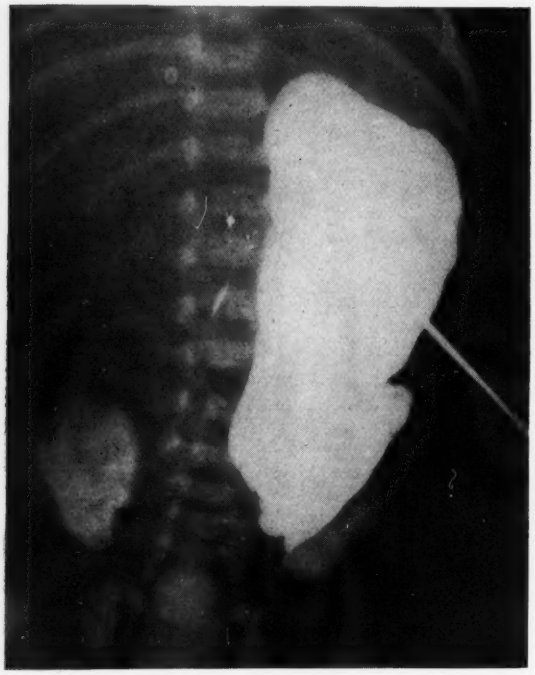


FIG. 338.—Note the strict unilateral localization of the barium in the perinephric space and in the retroperitoneal tissues below the kidney, except for the bulge in the lower lumbar region about the lower end of the mesentery. Injection pressure 45 mm. Hg.

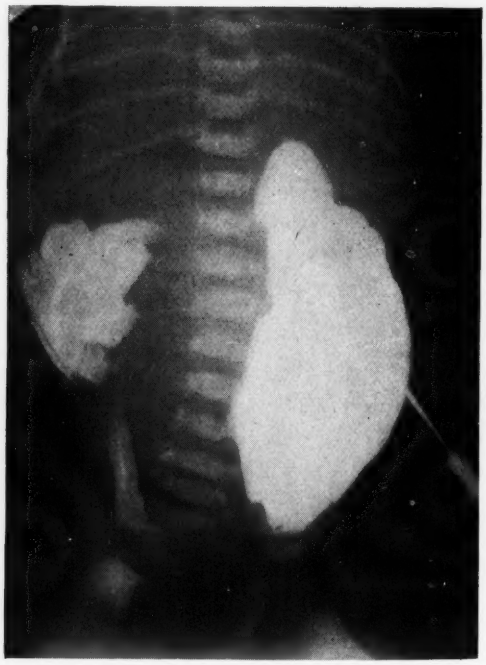


FIG. 339.—This radiograph shows the same features as *Fig.* 338. The irregular shadow on the opposite side was cast by barium injected into the paranephric space.

liable to displacement in the erect position, that even a perforate septum beneath it may provide reasonable support, and because its shorter vessels and nerves provide a good anchorage. Above the

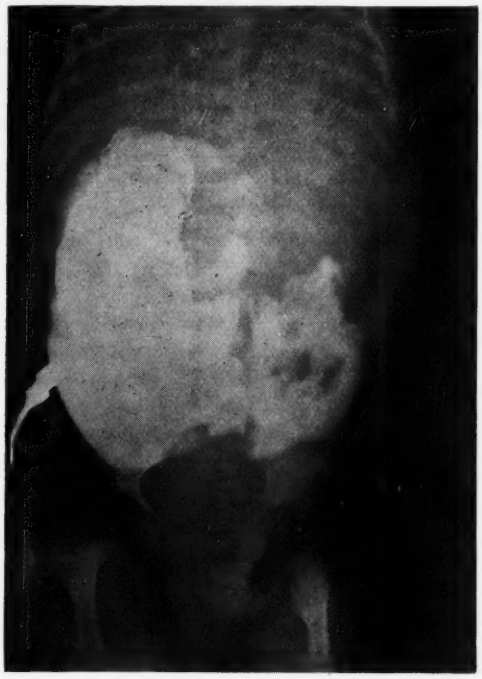


FIG. 340.—The barium has burst through to the opposite side in the lumbosacral region and has spread to involve the iliac and lumbar retroperitoneal regions of the opposite side. The pressure at this stage was about 105 mm. Hg.

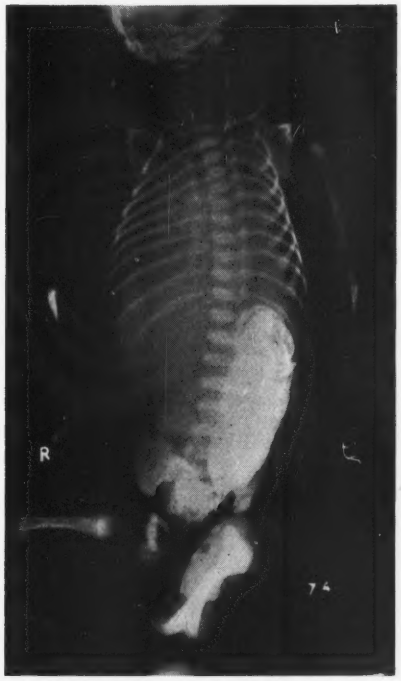


FIG. 341.—In this infant spread to the opposite side occurred through the base of the pelvic mesocolon. Observe the unilateral localization of the barium except at the point of rupture. Injection pressure at time of break through about 110 mm. Hg.

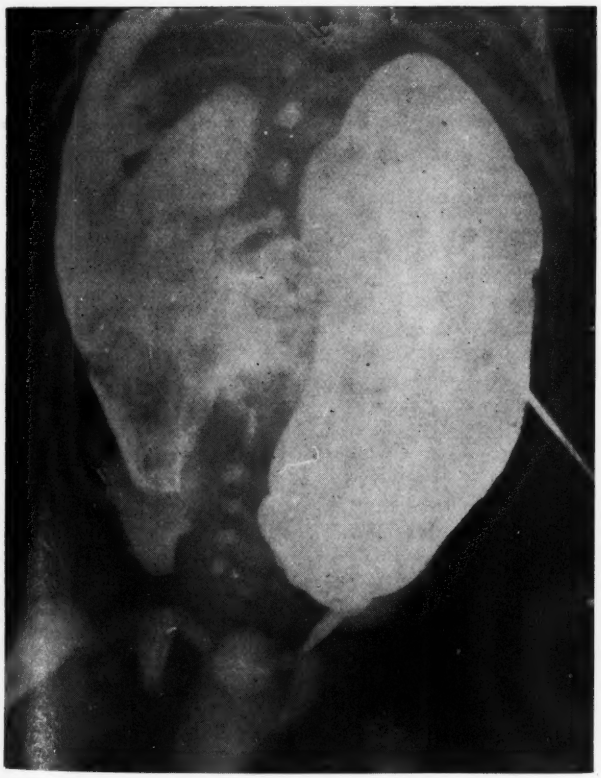


FIG. 342.—Rupture to the opposite side has occurred in the hilar region and the peritoneal space has been invaded. The pressure when rupture took place in this specimen was 130 mm. Hg.

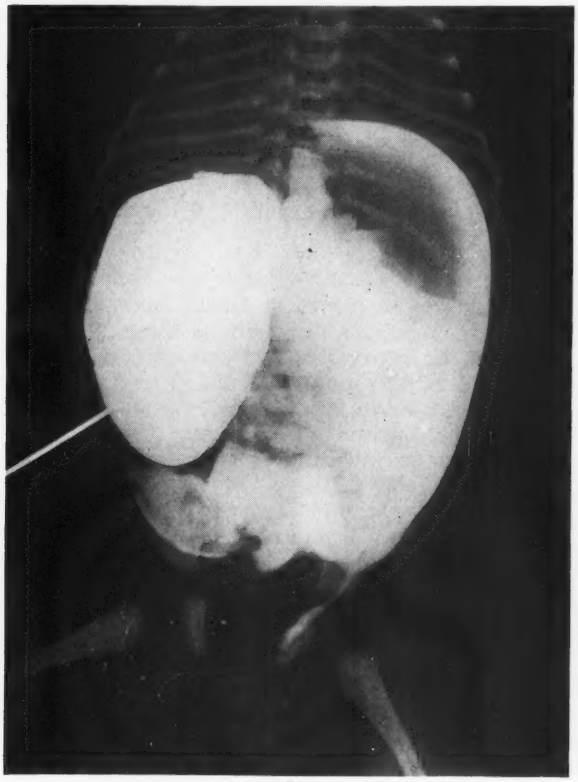


FIG. 343.—Rupture from the perinephric space has occurred in the suprahilar region, in this specimen with invasion of the peritoneal cavity. Some barium has spread upwards into the thorax alongside the aorta. There was no manometer in circuit when this picture was obtained, so the exact pressure cannot be given.

suprarenal the spread of the injection is limited by fusion of the two layers of renal fascia and their attachment to the diaphragmatic fascia (*Figs.* 331–333).

If the pressure is kept low the first sign of escape of fluid from the perinephric space is in the vicinity of the ureter (*Figs.* 332–335) and if the injection is continued very slowly the barium can then be seen spreading downwards and outwards, leaving a clear interval between it and the lower limit of the fluid within the perinephric space (*Figs.* 336, 337)—proof that except for a weak spot in the vicinity of the ureter, the two layers are fused inferiorly. This interval is quickly obliterated as the fluid spreads easily in the retroperitoneal tissues. If the fluid is injected rapidly the stage just described may be missed, or the two layers may be disrupted, giving rise to the erroneous impression that the renal fascia is like an open inverted sac. Zuckerkandl (1883), Sappey (1889), Gerota (1895), Gosset (1907), Southam (1923), Baumann (1945), and many others state that the two layers are not fused beneath the kidney, or form a funnel under it. Martin (1942) mentions that they are weakly fused inferiorly, and I believe this is correct and that the weakest spot is at the inferomedial angle of the perinephric space adjacent to the ureter.

Laterally the two layers are usually firmly united and are continuous with the retroperitoneal tissues, being also attached by short fibrous strands to the adjacent transversalis fascia. In several experiments the fluid escaped almost as easily on the lateral side as inferomedially, showing that Southam (1923) was not quite correct when he said that the union in this region is always very firm (*Fig.* 337).

Medially strict localization to one side was a constant finding until a late stage in the experiments (*Figs.* 338, 339) and the fluid which escaped from the perinephric space into the retroperitoneal tissues below the kidney also remained unilateral unless the pressure was raised by injecting more and more emulsion. Then the barium burst across to the opposite side below or through the lower end of the mesentery (*Fig.* 340), or even through the attachment of the pelvic mesocolon (*Fig.* 341), and invaded the corresponding pelvic, iliac, and lumbar retroperitoneal regions. In two cases when the injection pressure had risen to 160–170 mm. Hg the opposite perinephric space was invaded by upward spread of the fluid.

FIG. 344.—A transverse section of the infant whose radiograph is reproduced in *Fig.* 342, showing that the anterior layer of renal fascia and the overlying peritoneum have ruptured, with consequent involvement of the peritoneal cavity. This is the usual appearance when rupture occurs in the hilar region.

In 44 (69 per cent) of the 64 valid experiments extension to the opposite side occurred in the lumbosacral region: in 18 (28 per cent) rupture first occurred opposite or just below the renal hilum (*Fig.* 342), and in only 2 (3 per cent) did spread to the opposite side occur in the upper renal or suprarenal regions (*Fig.* 343). When rupture occurred near the renal hilum, in the majority (15 of the 18 cases) the fluid did *not* escape between the layers of the renal fascia, but burst through the anterior layer and the attached peritoneum, with consequent involvement of the peritoneal cavity (*Fig.* 344). In the remaining three specimens the barium infiltrated into the root of the mesentery. It will be evident that the fascia ruptured in the area where, during development, it is not reinforced by the dorsal mesogastrium or primitive mesocolon. The peritoneal cavity was involved only in one case when break through to the opposite side occurred in the lumbosacral or suprahilar regions (*Fig.* 343).

These experimental findings do not support the common belief that the two layers of renal fascia are fused medially only above the renal hilum.

On the medial side the two layers of fascia merge into the mass of connective tissue surrounding the great vessels, which is particularly evident in the region of the cœliac axis, superior mesenteric artery, and cœliac (solar) plexus (*Fig.* 345). Where the kidneys are in contact with the pancreas, duodenum, and colon the renal fascia also blends with the

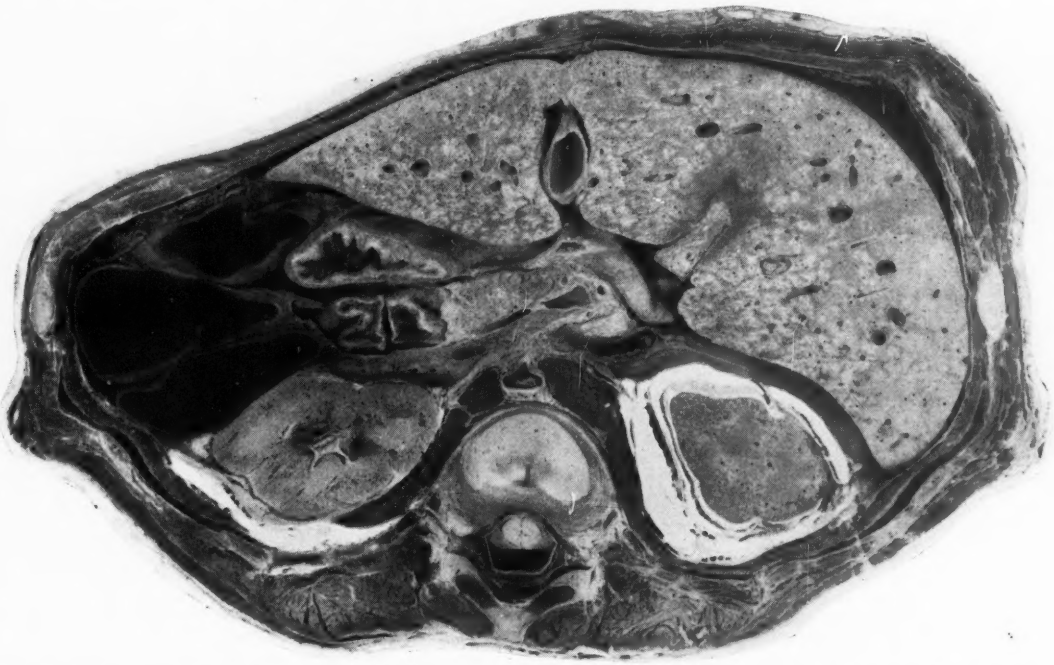


FIG. 345.—A transverse section at the level of the intervertebral disk between the last thoracic and first lumbar vertebræ. The right perinephric and left paranephric spaces have been injected with barium to show the limits of the spaces. On the medial side the two layers of renal fascia blend with the connective tissue around the aorta and inferior vena cava and distinct layers of fascia either anterior or posterior to the vessels cannot be distinguished.

FIG. 346.—A transverse section from the same specimen at the level of the disk between the second and third lumbar vertebræ. Note the unilateral localization of the barium and how the two layers of renal fascia merge into the connective tissue surrounding the great vessels. Where the anterior layer of renal fascia is in contact with the colon, duodenum, and pancreas it blends with the connective tissue behind them. The statement is commonly made that fat is absent in the perinephric space before the age of 10 years, but this is wrong; note, for example, the considerable deposit of fat in the left perinephric space of this stillborn infant; the fat in the right space is obscured by the presence of barium.

intervening layers of connective tissue (*Fig.* 346) and with the fascia in the root of the mesentery. The structures entering and leaving the kidneys penetrate this mass of connective tissue and are closely invested by it. Each posterior layer of renal fascia is partially blended with the fascia covering the

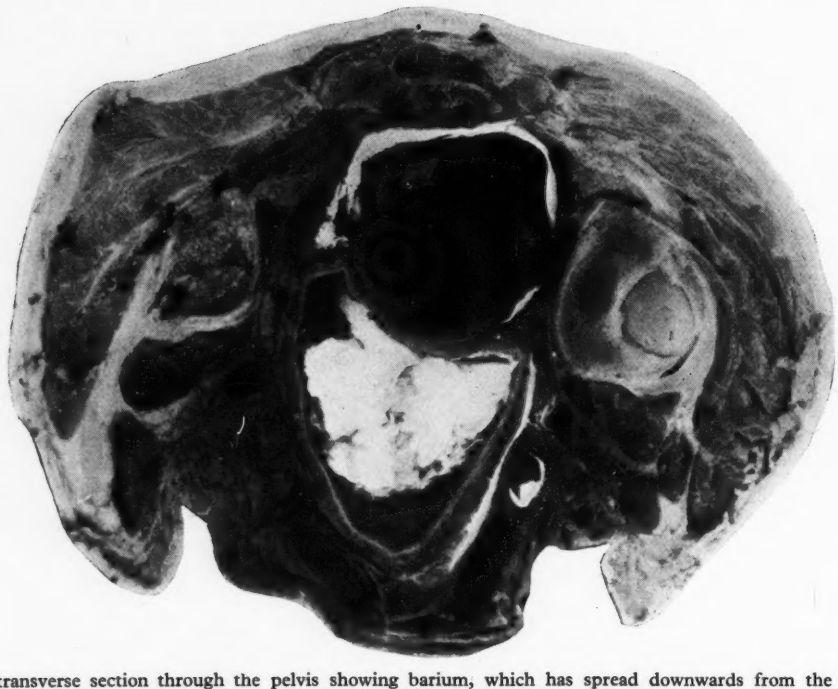


FIG. 347.—A transverse section through the pelvis showing barium, which has spread downwards from the perinephric space to invade the pararectal and paravesical regions. The barium within the bladder was injected directly in order to display its outlines more clearly.

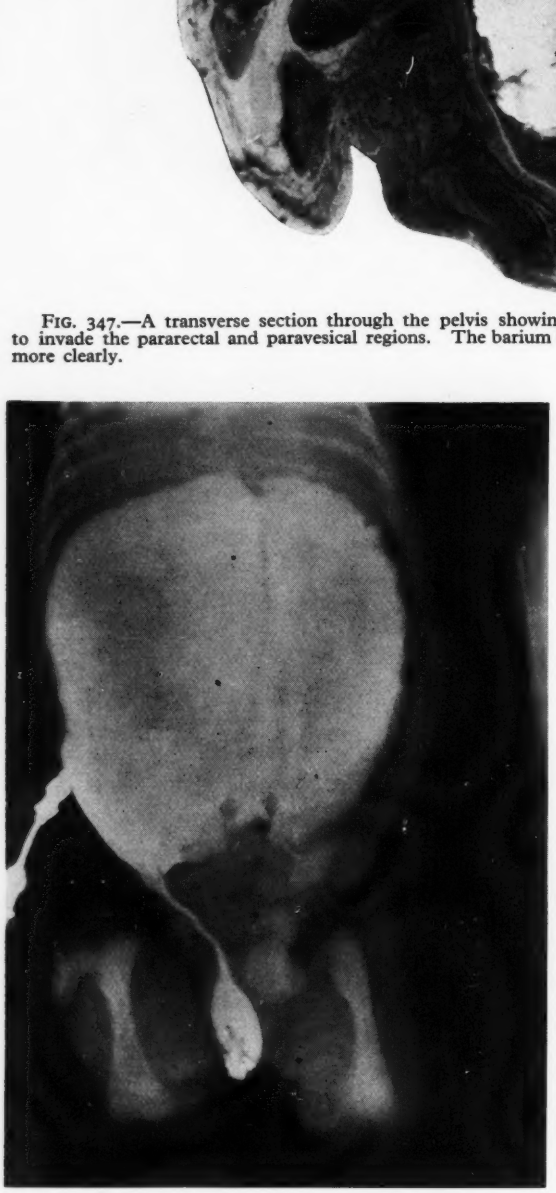


FIG. 348.—A specimen showing extension of barium in the extraperitoneal tissue surrounding a processus vaginalis into the scrotum.

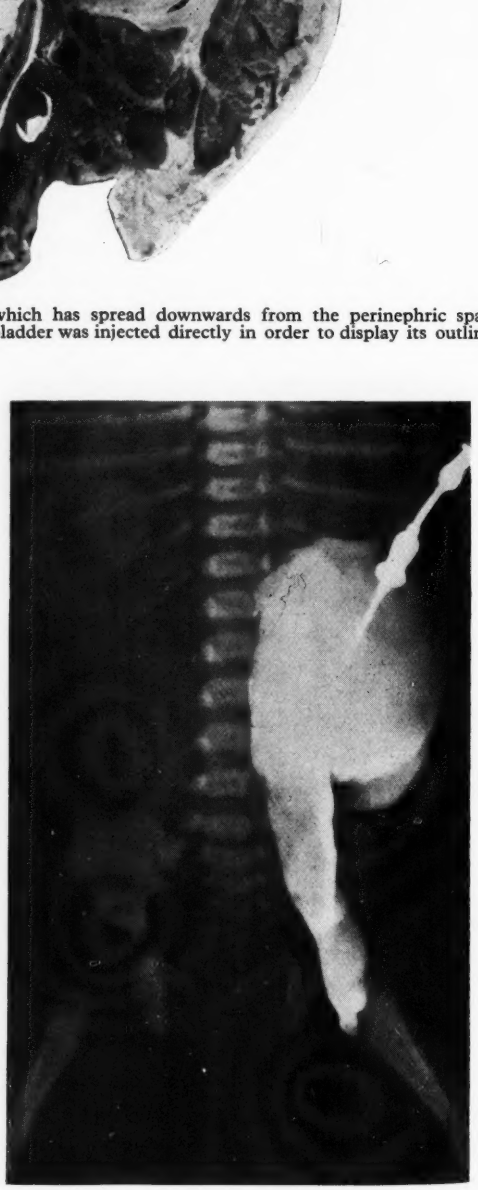


FIG. 349.—In this specimen the needle-point was displaced during an experiment and penetrated the psoas sheath. The barium passed easily downwards to the vicinity of the lesser trochanter on the same side.

corresponding psoas major and diaphragmatic crust, and through this connexion and their fusion with the connective tissue surrounding the great vessels both layers of renal fascia obtain an indirect attachment to the intervertebral disks and anterior spinal ligaments. Actually in the case of the posterior layer this attachment is more direct, as some of its fibres become fixed to the upper lumbar vertebræ and disks along the medial margin of origin of psoas major. The examination of increasing numbers of specimens of all ages has led me to believe that distinct layers of fascia, either anterior or posterior to the great abdominal vessels, can be created only by artificial dissection of the surrounding connective tissue.

The fact that the perivascular tissue is less dense below than above is shown by two of the experimental findings—spread to the opposite side was much more frequent in the lumbosacral region, and occasionally barium could be seen spreading upwards for a short distance in the perivascular tissues, particularly that surrounding the inferior vena cava (*Fig.* 340).

Extension into the pelvis in the loose retroperitoneal tissue was a common occurrence, with invasion of the paravesical and pararectal areas (*Fig.* 347), and sometimes the fluid spread through the inguinal canal around the processus vaginalis peritonei (*Fig.* 348), but, with one exception, there was no invasion of the thorax through the various openings in and behind the diaphragm even when the injection pressure was raised to 200 mm. Hg ; in the solitary exception barium passed upwards into the posterior mediastinum alongside the aorta (*Fig.* 343).

When the needle point was behind the posterior layer of renal fascia the fluid spread unevenly in the fluid spread into the pelvis and across to the opposite side, in a manner reminiscent of the spread from the perirenal space. This experimental finding tends to support the views of those clinicians who

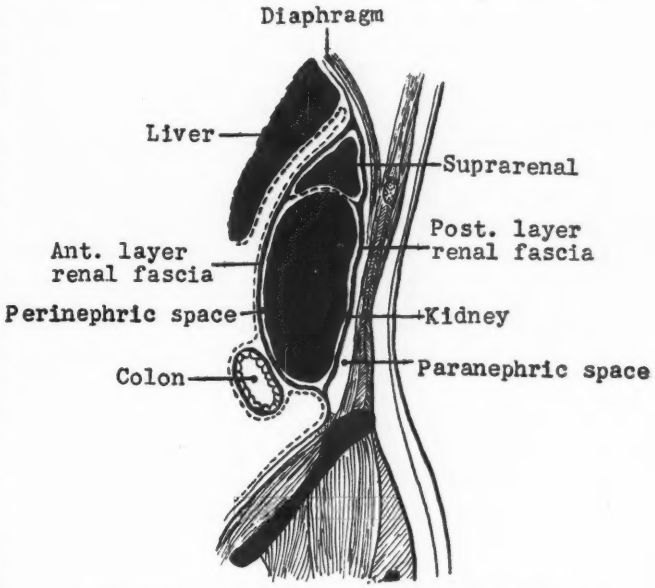


FIG. 350.—A diagrammatic representation of the renal fascia in a vertical section. The fascia is shown by a heavy black line.

believe that many 'perinephric' abscesses are in fact paranephric, i.e., they originate in the paranephric fat postero-inferior to the posterior layer of renal fascia, and seldom extend below the level of the iliac crest.

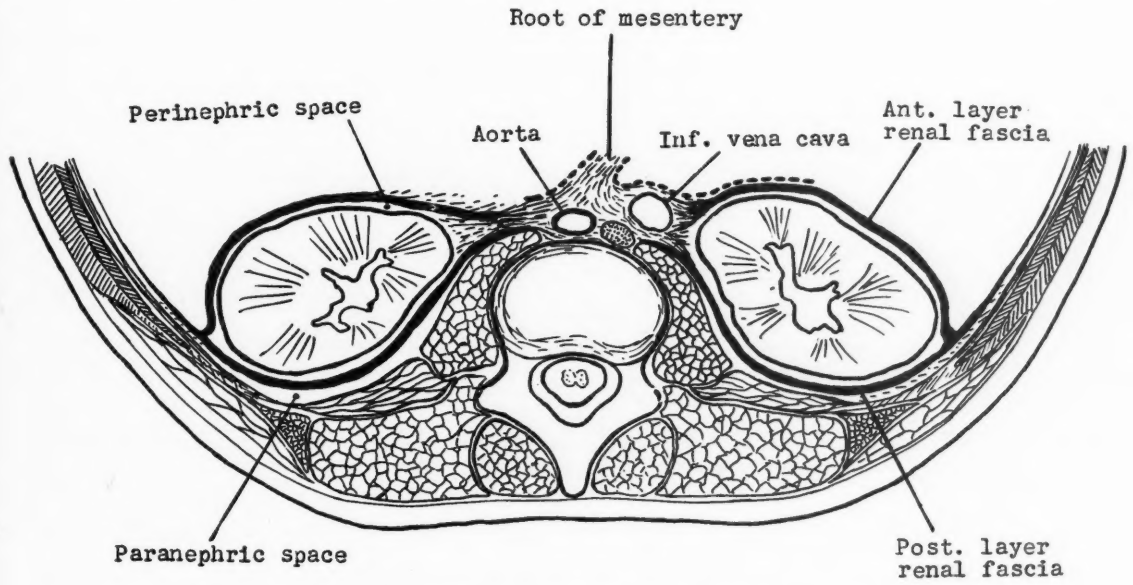


FIG. 351.—A diagrammatic representation of the renal fascia in a horizontal section.

front of the transversus, quadratus lumborum, and psoas major (*Figs.* 339, 345, 346) and downwards as far as the iliac crest, where it was often arrested as if there was some attachment of the retroperitoneal tissues in this region. If the pressure was then raised

In one experiment the needle point was displaced accidentally and penetrated the psoas sheath. The barium injection passed easily downwards to the thigh in the vicinity of the lesser trochanter (*Fig.* 349). This manœuvre was deliberately repeated in

three other infants without obtaining such good results, but it is interesting to record that in only one of the four did barium pass above the level of the medial arcuate ligament (medial lumbocostal arch) into the posterior mediastinum.

A diagrammatic representation of the arrangement of the renal fascia in vertical and horizontal sections is provided in *Figs.* 350, 351.

SUMMARY

1. Current descriptions of the renal fascia are more or less inaccurate.

2. The true disposition may be determined by routine anatomical procedures and by fluoroscopy and radiography during the injection of a radio-opaque substance.

3. The renal fascia is formed by a splitting of the retroperitoneal tissues to form a loose common investment for the kidney and suprerenal and, with few exceptions, there is no complete septum between the two organs.

4. The anterior and posterior layers of renal fascia are fused superiorly and laterally. Contrary to the common view, they are also united medially and inferiorly.

5. Above the suprarenal the fusion is firm, but the fusion of the layers beneath the kidney is weak, and the weakest spot, through which perinephric injections or effusions escape most easily, is in the vicinity of the ureter.

6. Medially the two layers blend with the mass of connective tissue surrounding the great vessels, in the root of the mesentery, and behind the pancreas and duodenum. Laterally they fuse together directly.

7. The two perinephric spaces are neither in actual nor potential communication, as the mass of connective tissue around the vessels, behind the pancreas and duodenum, in the upper part of the mesenteric root and in front of the vertebral column, presents an efficient barrier.

8. Direct continuity between the anterior and posterior layers of renal fascia of the two sides can be demonstrated only by artificial dissection.

9. Fluids escaping from the perinephric space spread downwards in the retroperitoneal tissue and may invade the pelvis, but until the pressure is raised by the injection of considerable amounts of fluid the spread is strictly unilateral.

10. Results are quoted showing that extension to the opposite side occurs most often in the lumbo-sacral region and is infrequent above the level of the renal hilum.

11. When rupture occurs in the hilar region, usually the anterior layer of fascia and the attached peritoneum both give way, and this permits invasion of the peritoneal cavity.

12. Spread upwards into the thorax through any of the openings in or behind the diaphragm is a rare occurrence, but extension in the extraperitoneal tissue around the processus vaginalis peritonei is not uncommon.

13. The experiments suggest that effusions spreading from the renal region which become arrested about the iliac crest are probably paranephric rather than perinephric in origin.

14. The spread of injections within the psoas sheath in a small group of cases in briefly described.

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REFERENCES

BAUMANN, J. A. (1945), *Acta Anat.*, **1**, 15.
BERGMANN, E. VON (1904), *A System of Practical Surgery*, **5**, IX. London: Williams & Norgate.
BILLINGTON. W. (1929), *Movable Kidney*. London: Cassell & Co.
GEROTA, D. (1895), *Arch. Anat. Entwck., Lpz.*, 265.
GOSSET, A. (1907), *Traité d'Anatomie Humaine* (Poirier and Charpy), **5**, I. Paris: Masson et Cie.
KNIGHT, G. D. (1893), *Movable Kidney and Intermitting Hydronephrosis*. London: Baillière, Tindall & Cox.
MARTIN, C. P. (1942), *J. Anat.*, **77**, 101.
MITCHELL, G. A. G. (1939), *Brit. med. J.*, **2**, 1134.
POIRIER, P. (1904), quoted by GOSSET.
SAPPEY, P. C. (1889), *Traité d'Anatomie Descriptive*, **4**. Paris: Delahaye et Lecrosnier.
SOUTHAM, A. H. (1923), *Quart. J. Med.*, **16**, 283.
STROMBERG, H. (1910), *Folia urol., Lpz.*, **4**, 533.
TOBIN, C. E. (1944), *Anat. Rec.*, **89**, 295.
VECCHI, A. (1910), *Anat. Anz.*, **36**, 149. (Good bibliography.)
ZUCKERKANDL, E. (1883), *Wien. med. Jahrb.*, 59.

ENGELMANN'S DISEASE

OSTEOPATHIA HYPEROSTOTICA (SCLEROTISANS) MULTIPLEX INFANTILIS; PROGRESSIVE DIAPHYSIAL DYSPLASIA

BY A. C. BINGOLD, LEEDS

ENGELMANN'S disease is an extremely rare disorder characterized by malnutrition, undervelopment, fusiform expansion and sclerosis of the diaphyses of the long bones of the limbs.

Credit for first drawing attention to this disease must be given to Engelmann, who published in 1929 "Ein Fall von Osteopathia hyperostotica (sclerotisans) multiplex infantilis". Without knowledge of Engelmann's paper Riley and Shwachman (1943) reported two patients with an identical dystrophy which they termed: "Unusual Osseous Disease with Neurologic Changes." Sear (May, 1948) described a further case under the eponymous title of "Engelmann's Disease". Soon afterwards Neuhauser, Shwachman, Wittenborg, and Cohen (July, 1948) presented a detailed study of four

patients suffering from "Progressive Diaphysial Dyplasia", including and following up in this series the boy and girl already investigated by Riley and Shwachman. During the preparation of the author's paper Michaelis (1949) published another example of "Engelmann's Disease".

The purpose of this article is to report briefly the above seven cases, to add an eighth, and to discuss fully the clinical, radiological, and pathological features, differential diagnosis, treatment, and prognosis of this uncommon disease.

CASE REPORTS

Case 1 (Engelmann, 1929).—Boy, aged 8 years. Four years' history of minor pains in his legs and a waddling gait. Quiet, unintelligent, poorly-nourished child. Height 110 cm., weight 16 kg. Disproportion between the various parts of his body: Neck too short, limbs relatively long. Bones frail, musculature feeble. Thickening of shafts of long bones of upper limbs and femora. Slight varus deformities of tibiæ. Lumbar lordosis and flat feet. Patellar reflexes brisk. Spleen enlarged. Testicles too small.

Radiographs: Symmetrical elongation and slight curving of the long bones with thickening and sclerosis of a large portion of their diaphyses. Individual bones, however, not all affected to the same extent—in some, practically the entire shafts, in others, only the proximal portions of the shafts dense and expanded. Bone ends, epiphyses, and joints normal. Marked sclerosis of frontal bones and base of skull. Small sella turcica.

Blood examination: Mild anæmia (R.B.C. 4,360,000 per c. mm., Hb. 72 per cent) with a lymphocytosis of 40·5 per cent, a slight monocytosis, and eosinophilia.

Case 2 (Riley and Shwachman, 1943; Neuhauser and others, 1948).—Girl, aged 4 years 3 months. Waddling gait. Tired easily. Appetite poor. Significant findings: underweight, "gait of straight-legged, wide-base, waddling type"; hyperactive tendon-reflexes and inconstant ankle-clonus.

Radiographs: Sclerosis of both clavicles and all the long bones of the extremities. Changes advanced in the long axis of the affected bones during the seven years patient was kept under observation.

Case 3 (Riley and Shwachman, 1943; Neuhauser and others, 1948).—Boy, aged 4 years 11 months. Chief complaint waddling gait since age of 3½ years. Always tired easily. Poor appetite. Well developed and well nourished, in contrast with previous and subsequent patients. Gait similar to that of *Case* 2. Inconstant ankle-clonus and hyperactive tendon reflexes.

Radiographs: Changes similar in distribution and character to those observed in Engelmann's patient, but frontal bone not involved.

Failed to put on weight. Weighed only 55·2 lb. when nearly 10 years old.

Case 4 (Sear, 1948).—Boy aged 10 years. Difficulty in walking since age of 3 years. Shaft of right femur tender just above adductor tubercle. Marked wasting of legs and some wasting of thighs. Mild degree of pes cavus and knock-knees. Knee-jerks and one ankle-jerk absent.

Radiographic changes the most extensive described so far: Sclerosis of the frontal bone, the first cervical vertebra, both innominate bones near the sacro-iliac synchondrosis, the ischia, the axillary borders of the scapulæ, the clavicles, and all the long bones of his arms and legs.

Case 5 (Neuhauser and others, 1948).—Girl aged 6 years. Gait waddling. Failed to put on weight in spite of good appetite. Diffuse atrophy of both lower limbs. Diminution of right patellar reflex.

Radiographic changes similar to those of *Case* 2.

Was followed up until she was 17 years and 4 months old. Menarche at 14 years was followed by dysmenorrhœa and oligomenorrhœa.

Case 6 (Neuhauser and others, 1948).—Girl, aged 4 years and 4 months. Abnormal gait since she began to walk. Vague muscular pains in her legs. Tired easily. Musculoskeletal system: Symmetrical generalized underdevelopment. No neurological abnormality.

Radiographs similar to those of *Case* 3.

Case 7 (Michaelis, 1949).—Female, aged 24 years. Complained of occasional ache in hip region and inability to stand, run, or kneel for long. History: Normal birth. Delayed first dentition. Aching in legs when 3 years old. At school excused games on account of weakness. Periods normal. Brother aged 32, limps; not examined. On examination: Above average intelligence. Under middle height, thin, small hands and feet, but long thighs. Bones of the extremities thickened and covered by poor muscles. Slight lumbar lordosis, knock-knees, valgus ankles. No neurological signs. Gait "resembles the uncertain gait of patients with muscular dystrophy". Moustache on her upper lip.

Radiographs: Distribution and character of changes as in Engelmann's patient.

Blood-picture: Slight anæmia with a monocytosis of 7 per cent.

Case 8 (Author's case).—This patient was referred to the Orthopædic Department of the General Infirmary at Leeds on Oct. 19, 1948 on account of vague pains in his left knee. He was subsequently admitted and fully investigated.

HISTORY.—He was 14 years of age. There is no evidence in his early history suggesting a future developmental aberration. The confinement was normal. He

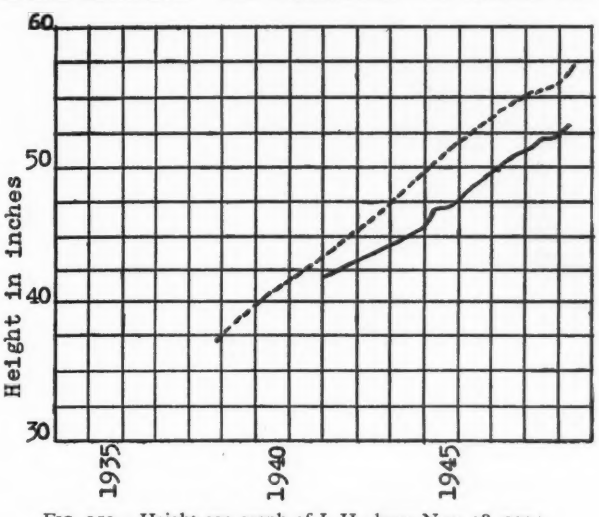


FIG. 352.—Height-age graph of J. H., born Nov. 18, 1934. Dotted line represents average growth of normal boys.

weighed 8 lb. at birth and measured 19 in. from head to foot. The first milestones were passed rapidly. At 4½ months he started saying his first words; his first teeth erupted at 6 months; when he was 9 months old he began to walk.

His appetite, however, has always been poor. After a few weeks of unsatisfactory breast feeding, artificial feeding was commenced. His diet was subsequently changed many times and always included liberal amounts of vitamin-D preparations, but he was never cured of his disinclination to eat.

When he was 7 years old his parents noticed that he was smaller than other children of corresponding age. How his rate of growth and increase in weight have lagged behind the normal is shown in *Figs.* 352, 353. Another example of his retarded development is provided by his delayed second dentition, his permanent upper incisors appearing at the age of 12 years.

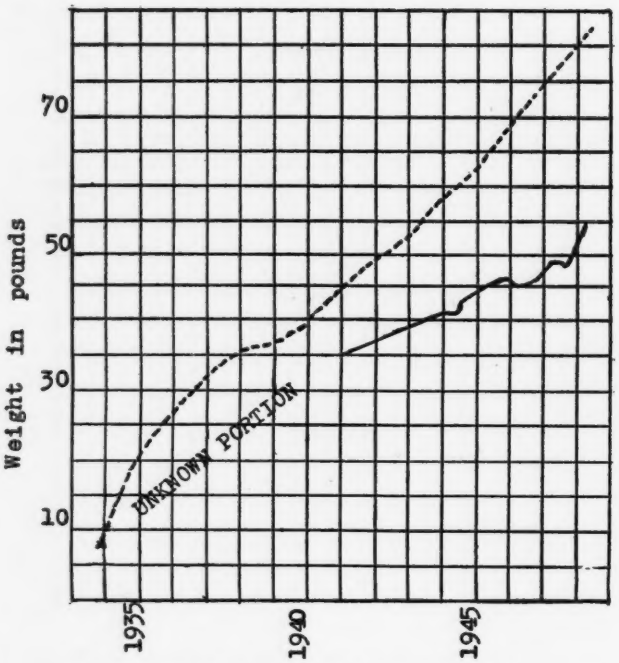


FIG. 353.—Weight-age graph of J. H. Dotted line represents average weight of normal boys. Notice the marked failure to gain weight. (Dotted curves in *Figs.* 352, 353, plotted from the data given by Moncrieff, 1948.)

He had measles when he was 1 year and diphtheria when he was 8 years old.

There is no evidence of any bone disease in his father, mother, younger brother, or any relations on either the paternal or maternal sides. His mother had no miscarriages.

PHYSICAL EXAMINATION.—The outstanding feature is the patient's poor physique. His height, weight, and measurements are well below par (*see Table* I).

Table I.—THE PATIENT'S HEIGHT, WEIGHT, AND MEASUREMENTS COMPARED WITH NORMAL VALUES

		Normal Values for a Boy aged 14 (After Holt and McIntosh, 1939)
Height	52 in.	58·6–62·8 in.
Weight	58 lb.	81·1–102·9 lb.
Intercristal diameter	7·125 in.	8·9–10·6 in.
Ratio $\frac{\text{crown to symphysis}}{\text{toes to symphysis}}$ (showing the relative elongation of his lower limbs)	$= \frac{25}{27} = 0{\cdot}925$	0·97
Head circumference	20·125 in.	21·6 in.
Chest circumference	24 in.	29 in.

His muscles are weak and his posture is faulty (*Figs.* 354, 355).

His gait is waddling, swaying. The Trendelenburg test is negative, but the altered architecture of his legs and feet provides ample explanation: a 30° external rotation deformity of the tibiæ, an outward slope of the lower tibial epiphyses (demonstrated by radiography), valgus heels associated with eversion of the feet and hypermobile first metatarsals—all these structural changes combine to effect an abnormal gait. Like all patients with hypermobile first metatarsals, he has callosities along the outer border of the heel, under the heads of the middle three metatarsals and the pad of the great toe, and smooth skin under the first metatarsophalangeal joint. When asked to stand on tip-toe he raises the proximal phalanx of the great toe off the floor, so that only the distal phalanx is weight-bearing on the inner side of the foot. This distribution of the body-weight over the foot is identical with that seen in hallux rigidus, but this disorder could not be demonstrated in the patient's foot.

He has no axillary hair and only a scanty growth of pubic hair. There is some fluff in his pre-auricular region, but none on his chin and above his lips. His testicles are in the scrotum but are small, measuring $\frac{3}{4}$ by $\frac{1}{2}$ by $\frac{1}{2}$ in.

His nervous, cardiovascular, and respiratory systems, abdomen, and urine show nothing abnormal.

The cause of the pain in his left knee is not clear.

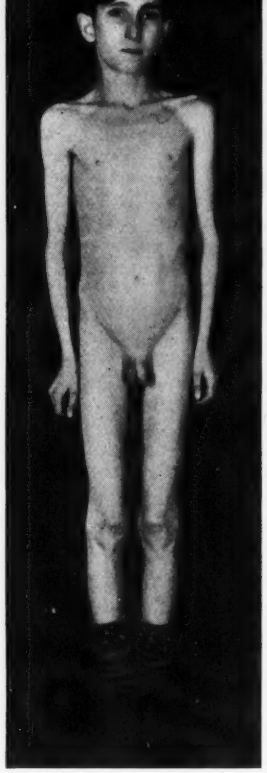


FIG. 354.—The patient from the front. Notice his thin, long arms and legs and his poor nutrition generally.

FIG. 355.—Posterior oblique view. Notice the lumbar lordosis and dorsal kyphosis.

RADIOGRAPHIC FEATURES.—The diagnosis of Engelmann's disease was made on the radiographic appearance of the patient's femora. I am indebted to Dr. Illingworth for the following report:—

Review of Bone Changes in Skeleton (*Figs.* 356–359).—"The bone changes in this case are mainly confined to the long bones of the lower limbs. The skull, spine, chest, clavicles, and upper limbs, including the wrists and hands, have a normal appearance. The pelvis is small, but otherwise does not show any abnormality.

"The bone changes in the femora are bilateral and symmetrical. Each shaft is slender and shows a relative

elongation. There is some coxa valga deformity, but the main changes lie in the distal part of the middle third of the shaft. In this region there is a definite expansion of the shaft and a considerable thickening of the corticalis. The bony thickening is both subperiosteal and endosteal, so that the medullary cavity is encroached upon. The transition from abnormal to normal cortex takes place fairly suddenly.

FIG. 356.—Radiograph of the upper three-fourths of the patient's femora.

"Just distal to the lesion, the finer trabeculæ of the spongiosa are ill defined or absent, but there appears to be some increased trabeculation in the distal part of the shaft.

"Growth lines are quite prominent.

"Similar changes to those described in the femora are present in both tibiæ, but rather less prominently.

"The condition would appear to be a bone dystrophy known under the names of 'Engelmann's disease' or 'progressive diaphysial dysplasia'. The appearances do not really suggest syphilis, but, nevertheless, this should be exluded."

The radiographs of the patient's wrists show six well-developed carpal bones but only the ossific nucleus of the pisiform. This represents a delay of four years in the appearance of this nucleus and is evidence of a skeletal maturation age of 10 years.

His dentition is also retarded, as is shown by the following dental formulæ, kindly supplied by Professor Read :—

Dentition: 6–4321/123456
6E4321/1234E6
Teeth unerupted: 87/78
875/578
Teeth missing: 5/ (this tooth might have been extracted)

5 appears normally at 10 years of age
7 appears normally at 12 years of age
8 appears normally over 20 years of age

LABORATORY INVESTIGATIONS.—

a. Biochemical :—

Calcium, Dec. 15, 1948, 10·3 mg. per 100 c.c. serum
Phosphorus, Dec. 15, 1948, 3·1 mg. per 100 c.c. serum.
Phosphatase, Dec. 15, 1948, 9·0 units.

FIG. 357.—Radiograph of lower two-thirds of the patient's femora. A different exposure was used to demonstrate the lines of arrested growth.

Calcium, phosphorus, nitrogen balance, Jan. 9, 1949–Jan. 22, 1949.

I am indebted to Dr. Dawson for the following report :—

Serum-calcium, 11·7 mg. per cent.
Diffusible calcium, 5·7 mg. per cent.
Phosphatase, 14 units per cent (normal 5–10 units per cent)

Balance Findings:—

	Ca. *mg./day*	*P.* *mg./day*	*N.* *gm./day*
Intake	338	1550	8·3
Urinary excretion	64	660	—
Fæcal excretion	295	486	—
Total excretion	359	1146	7·7
Balance	—21	+404	+0·6

"The patient is in calcium balance and has a marked positive phosphorus balance. A negligible positive nitrogen balance is also found.

"The retention of both calcium and nitrogen is less than I should have expected if active growth was occurring.

"The blood findings as far as calcium, phosphorus, and phosphatase are concerned are within normal limits. It must, therefore, be presumed that any abnormality found in the bones is due to a defect in growth generally and not due to an external cause rendering calcification deficient."

In addition, the following tests and examinations were carried out: Renal function tests, plasma proteins including albumin/globulin ratio, blood-cholesterol, serum sodium and potassium, blood carotene and vitamin A, 17 ketosteroid excretion, water dilution test for Addison's disease, basal metabolic rate, oral and intravenous dextrose tolerance tests, fractional test-meal, and chemical examination of fæces. They all gave results within normal limits.

b. Hæmatological: His blood-picture and bleeding and clotting time were normal.

c. Serological: The Wassermann reaction and the Kahn test were negative.

Biopsy.—On Jan. 24, 1949, a portion of the expanded femoral cortex and half the upper fibular epiphysis on the right side were removed for biopsy. The femoral periosteum was slightly thickened, but it stripped normally. The appearance of the underlying cortex was normal to the eye, although it proved very hard and was difficult to cut with the osteotome. There were no special features about the fibular epiphysis.

I am indebted to Dr. Collins for the following pathological report:—

"*Biopsy of a Femur and Upper Fibula.*—

"*Naked eye:*—

"1. The larger piece is from the upper end of the fibula and includes a slice of the epiphysis and a portion of the metaphysis.

"2. The smaller piece, about 1 cm. square, is from the shaft of the femur and this has been sawn into two across the grain of the bone.

"*Histology*:

"1. The portions of both diaphysial and epiphysial bone here show no obvious abnormality. The fibular cortex is thin and, at the metaphysis, shows the normal combination of resorption and regeneration. There is considerable endosteal osteoblastic activity along the under-surface of the cortex and around the cancellous trabeculæ. The growth cartilage appears normal and the pre-ossification columns are well formed. Immediately beneath this, however, ossification proceeds with scarcely any intervening osteoid zone. True bone appears prematurely and the abundant osteoblasts seem to be engaged upon the production of bone rather than osteoid. The lines of ossification and capillary advance are regular. (*Fig.* 360.)

"2. Sections of this cortical bone have been cut in a plane slightly inclined to the transverse. This is very dense and compact bone with Haversian canals somewhat irregularly spaced. Some canals are very small and lie at the centre of wide concentric lamellar systems. Other canals are wide and contain vascular connective tissue. A notable feature here is the abundance of prominent endosteal osteoblasts. Osteoid margins are not visible. There is no sign of bone resorption by lacunar osteoclasts in these sections. Unfortunately periosteum is not included. The large canals referred to lead from the general marrow cavity and are present mainly in the deeper part of the cortex. This process of endosteal osteoblastosis, uncompensated by resorption, must be leading to obliteration of the marrow cavity,

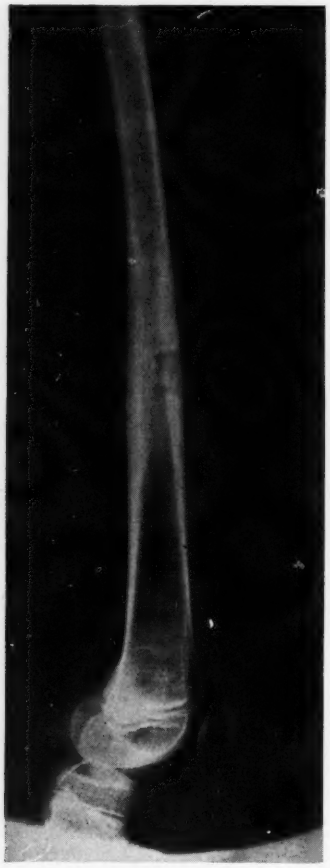
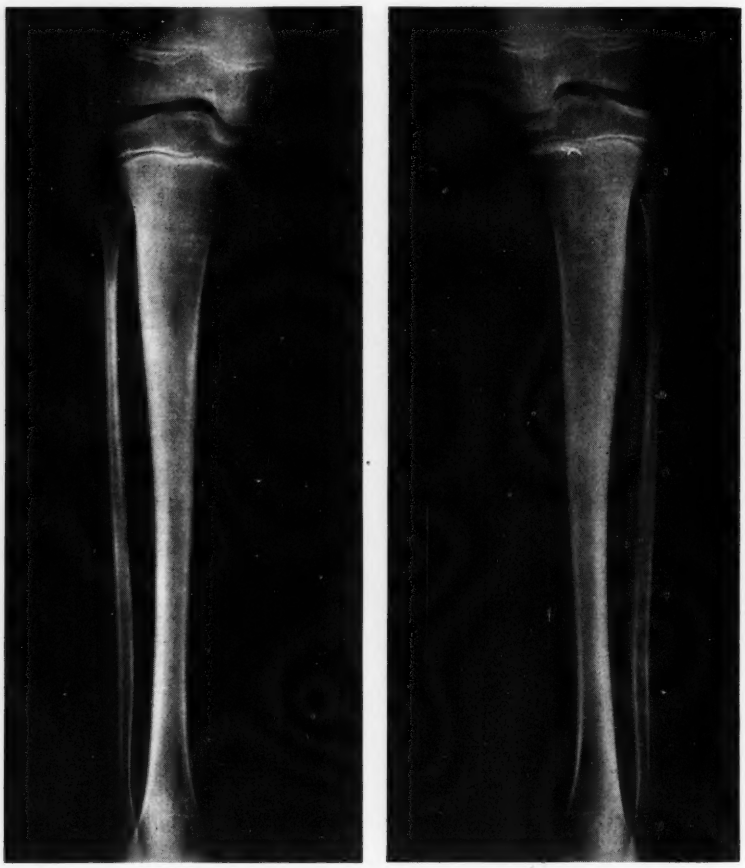


FIG. 358.—Lateral view of the right femur. The site of the biopsy is visible.



A B

FIG. 359.—Radiographs showing the patient's tibiæ.

although the process is doubtless very slow. Study of the bone fibrils by polarized light show a regular lamellar structure. This bone bears no resemblance to that of Paget's disease. (*Fig.* 361.)

"*In summary*: The microscopical examination of these biopsies shows: (*a*) prematurity of ossification at the upper growth cartilage of the fibula; (*b*) cortical hyperostosis, mainly endosteal, of the femoral diaphysis."

TREATMENT.—This was commenced on Jan. 21, 1949. To improve the patient's appetite the following mixture was prescribed: Tinct. Capsici min. 3, Mist. Gent. Alk. ad oz. ½., t.d.s., a.c. To accelerate his structural development intramuscular injections of 5 c.c. of Antuitrin Growth were ordered twice weekly.

FIG. 360.—Section of a portion of the patient's right upper fibular epiphysis.

FIG. 361.—Section of a portion of the sclerotic cortex of the right femur.

PROGRESS.—At the time of the writing of this paper the patient has been under treatment for four months. In the first two months he gained 6 lb. in weight and 1 in. in height. An attack of tonsillitis, however, halted this progress, so that there was no further improvement in the next two months.

DISCUSSION

Clinical Features.—

Sex and Age Incidence.—Of the 8 patients, 5 are male and 3 female. Their ages vary from 4 to 24 years.

Familial and hereditary tendencies have not been reported.

Symptoms and Signs.—The symptoms are as follows: Vague pains in the lower limbs, a peculiar gait, easy fatiguability, a poor appetite, and failure to gain weight and grow at the normal rate. Dentition may be delayed.

The stature of these patients is frail and abnormal. They are under-developed and poorly nourished. Their extremities are thinly clothed with weak muscles, through which the thickened shafts of the bones may be felt. Their arms and legs are relatively elongated, but the neck may be short. Additional, though inconstant, findings are a faulty posture, knock-knees, and flat-feet. In the author's patient flat-feet and hypermobile metatarsals are associated with an external rotation deformity of the tibiæ. Sear's patient differs from the others in having a mild degree of pes cavus.

The nervous system may show minor abnormalities: increased tendon reflexes were reported in 3 patients, diminished reflexes in 2, and only 3 are neurologically normal.

The peculiar gait, variously described as 'waddling', 'swaying', 'straight-legged, wide based, foot slapping', 'resembling that of a patient with muscular dystrophy', may be caused by neurological disturbances. However, in the author's patient it results from a combination of clear-cut abnormalities, which have already been described, and an unusual passage of the body-weight over the foot.

Hypogonadism has been noted in male and female patients: two, boys, including the author's patient, have small testicles. One girl started menstruating at 14 years of age and subsequently developed dysmenorrhœa and oligomenorrhœa.

The secondary sexual characteristics may also be affected. Thus the author's patient at 14 years of age had hardly any growth of facial, pubic, or axillary hair, but, by contrast, Michaelis's patient, a girl of 24, had a moustache on her upper lip.

Mild anæmia associated with a slight monocytosis was found in 2 patients. In Engelmann's original case this was combined with a lymphocytosis of 40·5 per cent and eosinophilia.

Intelligence is usually normal; only Engelmann's patient was described as unintelligent.

Pathology.—Microscopical examination of bone biopsies are recorded in *Cases* 3, 4, 6, and 8. It is

evident that the radiologically dense areas of the affected bones are areas of cortical hyperostosis. This increase in the compact bone of the shaft is due to new apposition of bone by osteoblastic activity on both periosteal and endosteal surfaces. Apart from its massiveness, the bone is of normal lamellar structure.

Sections in some instances have shown a combination of bone resorption and deposition. In *Case* 6 this combination of formation and resorption of bone was particularly seen in a biopsy taken from the transitional area between the normal and dense bone. Here there was also some fibrosis of the marrow. In general, however, marrow fibrosis is absent and it is to be noted that no cartilage is seen in the affected tissues. The abnormal appearance of the epiphysis in *Case* 8 probably represents a zone of arrested growth. No deductions as to aetiology can be made from the pathological changes which have so far been discovered in the affected bone, since these have no particularly specific character. Neuhauser and others (1948) state that the microscopical appearances are similar to those found in infantile cortical hyperostosis. Of more importance, however, is the absence of pathological features of certain other bone diseases, e.g., fibrous dysplasia, Paget's disease, Ollier's disease, osteomyelitis, and bone abscess.

The aetiology of the disease is completely unknown. Syphilis is not a factor. Biochemical studies have revealed no abnormality.

Radiological Features.—The radiological changes are diagnostic. They consist mainly of a symmetrical, dense, fusiform expansion of the cortex of the diaphyses of the long bones, tending to obliterate the medullary cavity. The transition from normal to abnormal bone is rather abrupt. The lesions, which are progressive in the long axis of the affected bones, have been observed in all the long bones of the body including the clavicles, but never in the short long bones of the hands or feet, or the ribs. In the author's case the changes are confined to the femora, where they are very obvious, and the tibiæ, where they are less well marked. The limbs are elongated relative to the size of the children.

Increased density has also been described in the frontal bones, the base of the skull, the first cervical vertebra, both innominate bones near the sacro-iliac synchondrosis, the ischia, and the axillary borders of the scapulæ.

In the X-ray films the epiphyses are normal, but there may be delay in their ossification.

Differential Diagnosis.—Before the diagnosis of Engelmann's disease can be entertained, every cause of dwarfism and infantilism must be ruled out. The final diagnosis depends upon the typical fusiform sclerosis of the shafts of the long bones as seen in X-ray films. Therefore, only those generalized diseases which are characterized by similar sclerosis merit discussion in the differential diagnosis. They are all excessively rare. *Fig.* 362 shows in diagrammatic form the bone changes of three conditions which may be confused with Engelmann's disease: (1) Osteopetrosis; (2) Infantile cortical hyperostosis; (3) Metaphysial dysplasia.

Osteopetrosis is too well known to require detailed discussion. Radiologically the affected bones are dense. The density tends to be most marked in the metaphysial regions and often presents a banded appearance. As a rule the ends of the bones are expanded.

The *Infantile Cortical Hyperostosis* of Caffey and Silverman (1945) is characterized by irritability, a mild infection of the upper respiratory tract, pleural exudates, fever, anæmia with leukocytosis, a subacute course, and sometimes recurrences. It has been observed in infants and children up to 3 years of age. The mandibles, the bones of the shoulder girdles, the bones of extremities, and the ribs may be attacked, but the distribution of the changes is not symmetrical. Clinically there are brawny, tender, soft-tissue swellings over the affected bones. Radiologically there is a periosteal reaction which is most intense at the mid-shaft. In recent cases it consists of a single line, but in older lesions it assumes a massive lamellated appearance. The cortex may become dense and expanded and may obliterate the medullary cavity. The width of the bone is apt to increase, so that its shape tends to become square. Within two to three years the affected bones become radiologically normal.

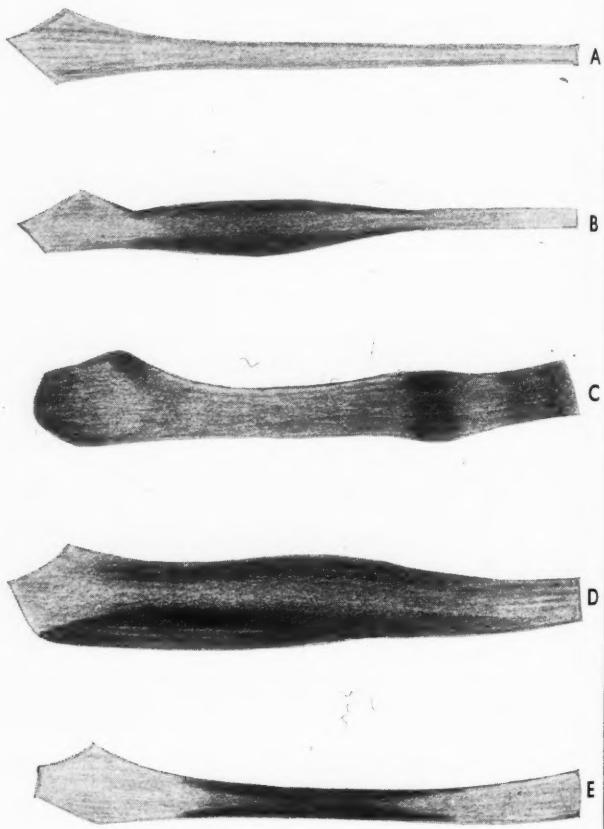


FIG. 362.—The ulna (in semidiagrammatic form). A, Of a normal person; B, In Engelmann's disease; C, In osteopetrosis; D, In infantile cortical hyperostosis; E, In metaphysial dysplasia.

It is interesting to recall that Hirsch (1929) published a case which in the author's opinion belongs to this group, although Michaelis (1949) has suggested that it be classed with Engelmann's disease.

Familial Metaphysial Dysplasia was first described under this name by Bakwin and Krida in 1937. However, Pyle (1931) has previously drawn attention to a similar case.

The occurrence of the disorder in two siblings suggests a hereditary factor.

The patients are tall, though frail, and have knock-knees and weak feet. The upper ends of the humeri and the ends of the femora and tibiæ about the knees feel expanded. There is limitation of movements at the elbows. The thighs and legs are unusually long.

Radiography shows that the changes in the bones are symmetrical. There is marked expansion associated with osteoporosis of the distal halves of the femora, the proximal thirds and distal thirds of the tibiæ and fibulæ, the proximal two-thirds of the humeri, the distal halves of the radi and ulnæ, the bones of the hands, the clavicles, and probably also the ribs. The middle portions of the shafts of the affected bones show sclerosis. The femora and tibiæ are abnormally long.

Increased radiographic density of the tubular bones is also a feature of the following affections :—

"*Dysostosis enchondralis metaphysaria*" was the name given by Murk Jansen (1934) to a unique skeletal disorder. The patient was a grossly deformed dwarf. Radiologically his long bones were irregular and abnormally dense. This applied particularly to the short long bones. The metaphyses were expanded and there were abnormally wide gaps between the epiphyses and the shafts which were occupied by numerous discrete centres of ossification. Haessler, quoted by Bernuth (1948), has described a similar disease in a six-year-old child. An injection of 7·5 mg. of vitamin D_3 was said to have led to prompt cure.

Congenital syphilis may cause a patchy, dense sclerosis and an expansion of the shafts of the long bones without formation of sequestra, rarefaction, or irregularity of the cortex. The changes, however, are asymmetrical and need not affect more than one or two bones.

Yaws may give rise to a similar radiographic appearance. Oosthuizen (1949) has published X-ray pictures of bones which bear a striking resemblance to Engelmann's disease.

Leukæmia, including myelosclerosis, may be associated with radiographic sclerosis of, and a periosteal reaction around, the shafts of the long bones. Blood and bone-marrow examinations may be required to establish the diagnosis.

A number of poisons have similar effects :—

Industrial fluorosis (Medical Research Council, 1949) is a system disease and attacks all the long bones of the body. In the cases studied so far this has not been associated with clinical disability. Radiography of the long bones demonstrates periosteal irregularities and sometimes exostosis. The spine exhibits marked lipping and calcification of its ligaments. The pelvis often has short exostoses projecting from the anterior iliac spines and there may be roughening of the iliac crests and the ischial spines. In a number of cases increased density of the bones has been observed.

Chronic lead, bismuth, and phosphorus poisoning may produce dense broad bands in the metaphyses.

Hypervitaminosis A (Toomey and Morisette, 1947; Rothman and Leon, 1948) is also a cause of cortical hyperostosis and periosteal reaction. The changes affect the skeleton symmetrically and are most intense at the mid-shafts. They resemble those seen in infantile cortical hyperostosis. However, the course of hypervitaminosis A is afebrile.

Treatment.—In the absence of specific therapy the undermentioned remedies may be tried during the growth period :—

The appetite of these patients may be improved by the stomachic bitters.

Their rate of growth may be accelerated by one of four treatments :—

1. Growth Hormone (Evans, 1935), sold under the trade name Antuitrin Growth by Park, Davis & Co.
2. Growth hormone and thyroid. (Bayer, 1944)
3. Chorionic gonadotropine. (Finkler et al. 1942)
4. Testosterone propionate. (Finkler et al. 1942)

If after a trial period of six months it has become obvious that the patient's height and weight have not increased, treatment by one of the above alternative methods should be begun.

Prognosis.—The late results of this progressive disease are unknown. Michaelis' patient, the oldest reported so far, is 24 years of age. However, the frailty of these patients suggests that they may die comparatively young from intercurrent infection.

SUMMARY

1. Engelmann's disease is characterized by malnutrition, undervelopment, and progressive hyperostosis and fusiform expansion of the cortex of the long bones. Sclerosis of the base of the skull, the first cervical vertebra, and portions of the pelvis has also been observed. Intelligence is usually normal.

2. To seven cases already published, an eighth is added.

3. The clinical, pathological, and radiological features, differential diagnosis, hormone therapy, and prognosis have been discussed.

My thanks are due to Mr. R. Broomhead, under whose care the patient was admitted, for encouraging me to write this paper; to Professor R. E. Tunbridge and Dr. E. C. Allibone for their generous help; to Professor T. Read for advising me regarding the patient's dentition; to Dr. G. H. Illingworth for the radiological report; to Dr. D. H. Collins for the pathological report; to Dr. J. Dawson for the calcium, phosphorus, and nitrogen balance findings; to Dr. H. G. Hutton for the patients' height-age and weight-age graphs.

REFERENCES

BAKWIN, H., and KRIDA, A. (1937), *Amer. J. Dis. Child.*, **53**, 1521.

BAYER, L. M. (1944), *J. clin. Endocrinol.*, **4**, 297.

BERMUTH, V. F. (1948), "Fiat Review of German Science, 1939–1946", *Pædiatrics, Wiesbaden*, 84.
CAFFEY, J., and SILVERMAN, W. A. (1945), *Amer. J. Roentgenol.*, **54**, 1.
ENGELMANN, G. (1929), *Fortschr. Röntgenstr.*, **39**, 1101.
EVANS, H. M. (1935), *J. Amer. med. Ass.*, **104**, 464.
FINKLER, R. S., FURST, N. J., and COHN, G. M. (1942), *J. clin. Endocrinol.*, **2**, 6.
HIRSCH, T. S. (1929), *Radiology*, **13**, 44 (*Figs.* 51–55, pp. 63–65).
HOLT, L. E., and MCINTOSH, R. (1939), *Holt's Diseases of Infancy and Childhood*, 23. New York and London.
JANSEN, MURK (1934), *Z. orthop. Chir.*, **61**, 253.
MEDICAL RESEARCH COUNCIL (1949), *Industrial Fluorosis*, Memorandum No. 22, London.
MICHAELIS, L. S. (1949), *Proc. R. Soc. Med.*, **42**, 27[illegible].
MONCRIEFF, A. (1948), *Hutchinson's Lectures on Diseases of Children*, **1**, 39. London.
NEUHAUSER, E. B. D., SHWACHMAN, H., WITTENBORG, M., and COHEN, J. (1948), *Radiology*, **51**, 11.
OOSTHUIZEN, S. F. (1949), *Brit. J. Radiol.*, **22**, 276.
PYLE, E. (1931), *J. Bone Jt Surg.*, **18**, 874.
RILEY, C. M., and SHWACHMAN, H. (1943), *Amer. J. Dis. Child.*, **66**, 150.
ROTHMAN, P. E., and LEON, E. E. (1948), *Radiology*, **51**, 368.
SEAR, H. R. (1948), *Brit. J. Radiol.*, **21**, 236.
SMYTH, F. S., POTTER, A., and SILVERMAN, W. (1946), *Amer. J. Dis. Child.*, **71**, 333.
TOOMEY, J. A., and MORISETTE, R. A. (1947), *Ibid.*, **73**, 473.

ADENOCARCINOMA OF THYROID GLAND

A CASE REPORT; AND DISCUSSION OF THE SIGNIFICANCE OF THE HÜRTHLE CELL

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To date there has been no report in the literature of a carcinoma of the thyroid gland, exhibiting papillary structure, in which the constituent cells have been large granular cells with eosinophilic cytoplasm. The following case is, therefore, worthy of recording.

CASE REPORT

A. L., female, aged 53, was admitted to King's College Hospital on April 22, 1948. She complained of multiple nodules in and under the skin of the neck in the region of a previous thyroidectomy scar.

She had first noticed a goitre in 1936 and it had increased in size gradually. Growth had been more rapid for one year preceding November, 1946. Apart from nervousness and jumpiness, and some degree of stridor, she was otherwise perfectly well.

On examination of her thyroid in November, 1946, she was found to have a single nodule just to the right of the midline of her neck. It was apparently pressing on her trachea.

Partial thyroidectomy was performed on Nov. 29, 1946, under the impression that the nodule was benign. From this operation the patient recovered completely, but three to four months after operation a swelling the size of a hazel-nut appeared at the lower anterior angle of the left posterior triangle. This was followed by the appearance of numerous other nodules as shown in the photograph of this patient (*Fig.* 363). These nodules were not attached to skin, and many of them were not attached to deep structures and were readily mobile. Some of the deeper ones were apparently adherent posteriorly and there was a hard mass palpable in the site of the thyroid gland.

On April 23, 1948, a small subcutaneous nodule was removed for section.

The patient was referred to the Royal Cancer Hospital for in-patient treatment by deep X-ray therapy. By July 16, 1948, she had received a dose, 3000 r max., 2100 r min., in twenty-seven days, given by H.V.T. 400 kv.

She was seen in September, 1948. The lumps in her neck were very slightly smaller, and were softer. There was little other change, and the patient's general condition was good.

Comment.—Malignancy of the thyroid gland was diagnosed from the specimen obtained at the partial thyroidectomy. The cut surface was rather mottled with grey and brown islands, and the central parts were soft and necrotic. The neoplastic process could

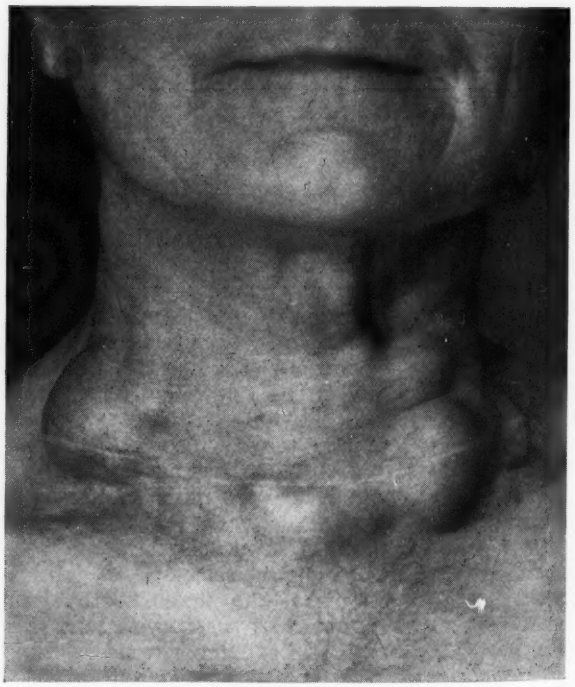

FIG. 363.—A photograph of the recurrences found in in the case here reported.

be seen originating from a nodule. A low-power microphotograph of the tumour is shown in *Fig.* 364. The greater part of the tumour exhibits papillary structure, though acinar arrangement is present in some areas, and in others the cells are arranged irregularly in sheets. The cells themselves are large, irregular in shape, and

they have a markedly eosinophilic and granular cytoplasm (*Fig.* 365).

The colour of the secondary deposit excised from the neck on cross-section was pinkish-white. Its structure is illustrated in *Fig.* 366. The cells are spheroidal, with eosinophilic and granular cytoplasm. Their nuclei are vesicular. The arrangement in this secondary growth is not predominantly papillary; sheets of cells are found,

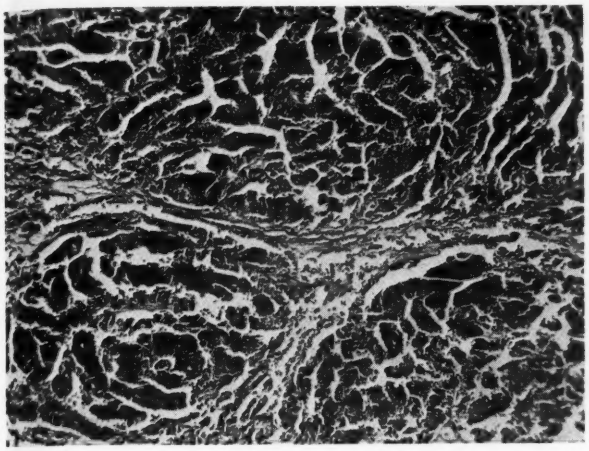

FIG. 364.—A low-power view of primary growth removed from the case reported in this article. H. and E. (× 20.)

columns are formed in some places, and occasional acinus formation is seen. There is a scanty vascular stroma.

DISCUSSION

In 1894 Hürthle described certain cells which he considered to be normal constituents of the thyroid, and to be capable of reproducing normal thyroid tissue when necessary. The cell now commonly called the Hürthle cell is large and polyhedral, 15 to 20 microns in diameter, rich in an eosinophilic and granular cytoplasm. The small nucleus is eccentrically placed, vesicular, and contains much chromatin clumped into nucleoli. Hürthle found such cells in the thyroid glands of puppies, and they were the same as the so-called parafollicular cells previously described to the Royal Society in 1876 and 1881 by Baber (Lennox, 1948). Noridez, Zechel, and Räymond (quoted by Lennox, 1948), have made detailed studies of these cells in dogs, cats, and rabbits.

Lennox (1948) pointed out that the relationship of these cells in animals to so-called Hürthle cells seen in adult thyroid glands was uncertain. Among histologists of repute there is little support for the occurrence of any similar cells in the normal human thyroid gland. Maximov and Bloom (1942), Means (1937), and others do not describe them. Lennox also noted that parafollicular cells were only frequently found in the first few weeks of life in those animals in which they occurred, whereas the large cells with eosinophilic and granular cytoplasm found in the human thyroids were generally found in the older age groups.

Askanazy, in 1898, was the first to describe these cells in toxic thyroid glands. For this reason, Lennox suggested that they should be called Askanazy cells. In his article (Lennox, 1948), he has shown the occurrence of such cells in various types of thyroid gland. In apparently normal thyroids they were absent in the younger age groups and uncommon in old men; they were more common in older women, and over the age of 60 were found in 50 per cent of the glands. Lennox noted a close relationship to lymphocytic hyperplasia. In glands

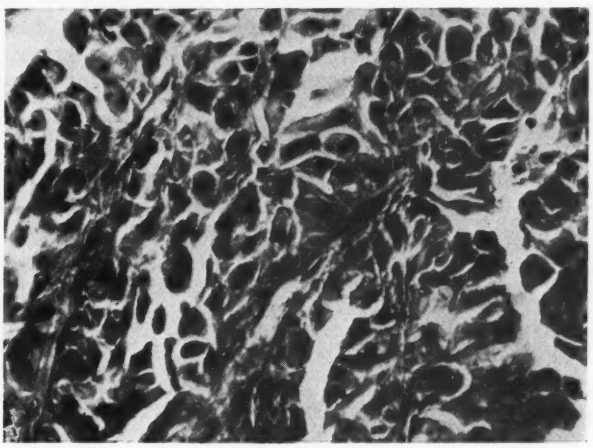

FIG. 365.—A high-power view of the growth illustrated in *Fig.* 364. The large cells shown here have a granular and eosinophilic cytoplasm, and are of the type described as Hürthle, or Askanazy, cells. H. and E. (× 300.)

from cases of thyrotoxicosis in women these cells were found more frequently than in normal glands of corresponding age groups; again there was an association with lymphoid hyperplasia. The incidence before the days of iodine was much the same,

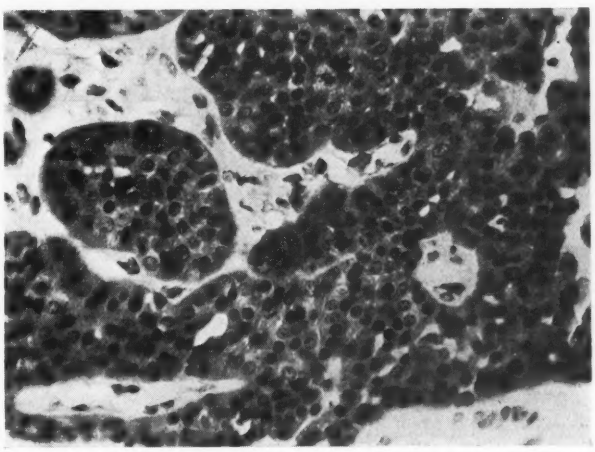

FIG. 366.—Sheets and columns of granular eosinophilic cells, with occasional acinar arrangement, can be seen in this high-power view of recurrence excised from the case reported. H. and E. (× 300.)

and thiouracil seemed to have had little effect on the occurrence of these cells. Lennox found no correlation between the presence of these so-called Askanazy cells and any clinical feature except sex and nodularity. In nodular goitres made up of nodules of non-proliferating and non-hyperplastic tissue these cells were infrequent. In cases of thyroiditis of Hashimoto type, Askanazy cells in

association with lymphoid hyperplasia were a marked feature. In the fibrotic Riedel's type these cells were not found.

Tumours of the thyroid gland made up of Hürthle cells have been called many names. Getzowa, who considered that masses formed of these cells originated from remnants of the glandular parenchyma of the postbranchial cell body, or lateral anlagen, called them ' postbranchial-cell tumours '. Langhans (1907) would appear to have subscribed to this theory of origin in so far as his alternative name to large-cell, small-alveolar, adenocarcinoma of the thyroid was ' parastruma postbranchialis '. Eisenberg and Wallerstein (1932), basing their theory on the resemblance to the oxyphil cells of the parathyroid gland, postulated that these tumours originated in cell rests of parathyroid tissue. By others, they were termed ' Hürthle-cell tumours ' (Martin and Elkin, 1939; Reimann, 1943).

Wilensky and Kaufmann (1938) recognized the mixed histological picture in their cases, and suggested that some change occurred in the cells of a nodule or carcinoma to produce an eosinophilic and granular cytoplasm. They did not consider that the behaviour of the tissue mass was related to the change in the cells, but to the nature of the precursor cells in which the change occurred. Lennox subscribed to this view, and postulated that it occurs in two main ways: by metaplasia in a nodule composed of proliferating tissue of fœtal type, and by metaplasia in what he called a ' toxic adenoma ', a nodule composed of hyperplastic tissue. In the latter case he considered that the hyperplastic thyroid acini may become ' worked out ' and pass through what he called a phase of Askanazy change and lymphocytic infiltration, before finally disappearing.

A similar theory might explain the appearances seen in Hashimoto's disease. Smith and others (1934) found increase in cell size and change to eosinophilic cytoplasm, in association with lymphoid hyperplasia, after compensatory hypertrophy and hyperplasia of the thyroid epithelium had failed to combat a lymphocytic infiltration. These authors made no mention of Hürthle cells, but they suggested that this change in the thyroid epithelium was a degeneration due perhaps to exhaustion of the cells, perhaps to ' choking ' by the lymphoid tissue.

When studying sections of nodules and of malignant growths of the thyroid, I was interested in the frequency of occasions on which I was able to identify relatively large cells, with granular eosinophilic cytoplasm, which so far as I could see were of the type called Hürthle cells. *Figs.* 367, 368 were obtained from a patient who was originally thought to have suffered and died from carcinoma of the thyroid gland. There is no doubt that he died from the effects of widespread carcinoma, but not of his thyroid gland. The thyroid itself appears necrotic, to some extent perhaps as a result of post-mortem change, and there is a diffuse lymphocytic infiltration. The remarkable thing about his gland, however, is the appearance of many of the acini and their epithelium. The acini are obviously in process of disintegration, and the lining epithelium is in some places broken away, in others still intact (*Figs.* 367, 368). It is composed of large eosinophilic cells, with a granular cytoplasm. In other areas the change is less marked. I wondered when looking at this specimen, whether the so-called Hürthle cells, or Askanazy cells, call them what you will, might be degenerating thyroid epithelium. With this idea in mind, I looked again at many sections. In specimens from 6 of the 22 cases of malignant disease of the thyroid which I have been able to study, there have been appreciable numbers of large cells with small vesicular nuclei with prominent nucleoli and granular and distinctly eosinophilic cytoplasm. Of these 6 cases, 3 are diffuse anaplastic adenocarcinomata. In all these the appearance of the cells resembling the so-called Hürthle cells is suggestive of a degenerative process occurring in otherwise relatively normal but small thyroid acini, perhaps as a result of invasion and as a step towards destruction by the neoplastic tissue. Sometimes the acinar arrangement is maintained, while in other places the acini appear to be disintegrating, with dispersal of the granular eosinophilic cells.

FIG. 367.—On the left is seen rather necrotic tissue found at post mortem in the thyroid of an old man of 66. Even at this relatively low power, acinar arrangement is easily recognized. On the right is an undegenerated nodule of thyroid tissue. H. and E. (× 70.)

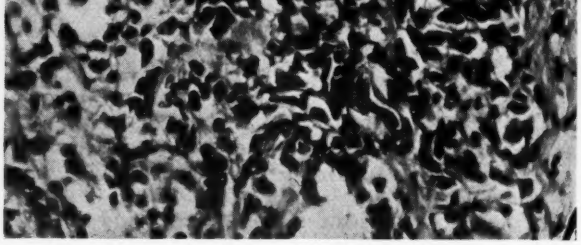

FIG. 368.—A high-power view of the degenerate thyroid gland shown in *Fig.* 367. The epithelium lining the disintegrating acini shows markedly eosinophilic cytoplasm. H. and E. (× 300.)

Figs. 369, 370 are of interest. These microphotographs are taken of a nodule, encapsulated by a relatively thin layer of compressed thyroid tissue, which was removed from the thyroid isthmus of a woman aged 48. The nodule was 2½ in. in diameter, and in the periphery it is composed of tissue of the fœtal type. In parts of the periphery there is acinus formation, and in a number of acini well-stained colloid is seen; in the periphery the cytoplasm of the cells, though in places somewhat granular, is definitely basophilic. The centre of the nodule is grossly degenerated, with massive

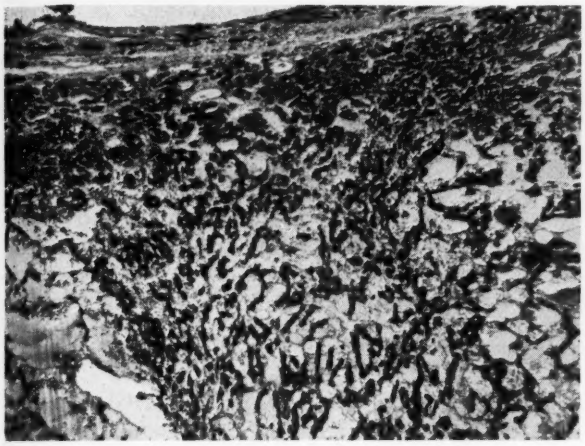

FIG. 369.—A view of a single discrete nodule removed from a woman aged 48. The capsule is poorly developed, and is made up of compressed thyroid tissue. The periphery of the nodule resembles the periphery of a nodule of fœtal type, but the centre is grossly degenerated, with large areas of massive hæmorrhage into loose stroma. Through the centre run strands of cells which become more eosinophilic as one passes inwards, cell outlines become more indistinct, and granularity of the cytoplasm becomes more marked. In the periphery there are some small primitive acini which contain well-stained colloid. There is no attempt at acinus formation centrally. H. and E. (× 20.)

hæmorrhages into a very loose stroma. Through this stroma stream columns of cells, appearing as though they have invaded it by proliferation from the periphery. As one passes in from the periphery towards the centre, the cell-type changes gradually but markedly. The cell outlines become more irregular and more indistinct, until, in the centre, the arrangement appears to be syncytial; and the cytoplasm of the cells merges almost imperceptibly from the somewhat granular and basophilic type peripherally into a definitely eosinophilic and markedly granular type centrally. The occurrence of syncytial masses in what they called a benign Hürthle-cell 'adenoma' of the thyroid was described by Eisenberg and Wallerstein; so perhaps it is not correct to call the nodule described above, malignant. It certainly seems that the granular eosinophilic cells in the centre are derived from the relatively normal type of young thyroid cells at the periphery. The gradation of cell type seen as one passes in toward the degenerated centre of the nodule is extremely suggestive that the change is a secondary one; one might postulate a degenerative process due to defective nutrition of the proliferating cells. According to Lennox, the change should be termed a metaplasia.

So we return to the case reported at the beginning of this article. There is no doubt of the malignancy of this tumour, for recurrences and secondary deposits developed after a partial thyroidectomy. Some of the tissue shows definite and clear-cut papillary structure, in some areas there are sheets of tumour cells, and in others there is acinar arrangement. The interest lies in the cell type, which is preponderantly granular and markedly eosinophilic in both papillary and non-papillary areas. In the papillary areas the appearance is suggestive of a secondary change having occurred in the cells (*Fig.* 365). The report on the macroscopical appearance of the tissue removed at the partial

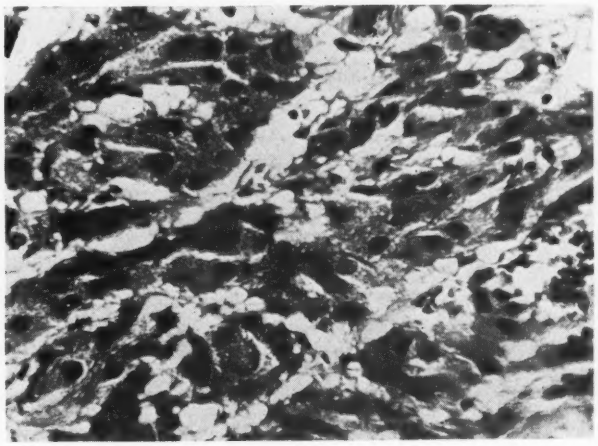

FIG. 370.—A high-power view near the centre of the nodule shown in *Fig.* 369. In this area the cells are definitely eosinophilic and the cytoplasm is obviously granular. The syncytial appearance, with indefinite cell outlines, can also be seen. H. and E. (× 300.)

thyroidectomy is also suggestive: "the cut surface was rather mottled with grey and brown islands, and the central parts were soft and necrotic."

From the material I have been able to study it is clear that the cells so long known as Hürthle cells originate from adult thyroid epithelium and from the young epithelium of nodules of fœtal type. These cells are found, as shown by Lennox, in thyroid glands in various diseases, in old age, and perhaps more commonly in women. They seem to become more obvious when thyroid acini and their epithelium are subject to disintegration, whether autolytic, or as a result of over-activity, or invasion by neoplastic tissue, or defective blood-supply. It would seem most likely that the change to cells with granular and eosinophilic cytoplasm is a form of degeneration which may occur in thyroid epithelium, whether benign or malignant, or in a primary or a secondary tumour. Lennox seems to have reached the same conclusion, because he said that the appearance of this type of cell must be regarded as a regressive phenomenon. He found no evidence that these cells could secrete thyroxin. In all areas made up of these cells which I have been able to identify, any acini formed by them have been devoid of colloid, and in the reports in the literature colloid formation has not been a prominent feature. These findings tend to confirm the view that they are non-functional, a view borne out by Rawson and others (1948) in their investigations of function of various thyroid nodules and tumours by 'take-up' of radio-active iodine.

It would seem reasonable to discard the term Hürthle cell as applied to nodules or to malignant tumours. It is unlikely that the cells under consideration are the cells Hürthle described. I have mentioned earlier that Wilensky and Kaufman considered that the important factor was the nature of the cell type in which the change occurred. Ewing (1940) considered that the course of malignant tumours composed of these cells was essentially the same as that of other tumours of the thyroid, and that treatment should be the same. Tumours showing cells of this type should be classified according to the nature of the precursor cells, and other characteristics normally used.

I wish to thank Mr. E. G. Muir for permitting me to report this case; Dr. B. Evans, Pathologist, Queen Victoria Hospital, East Grinstead, for sending me slides of the primary growth, and her report; Dr. Lederman, Assistant Radiotherapist, The Royal Cancer Hospital, for undertaking treatment of the patient; Professor H. A. Magnus, and his Department of Pathology; Dr. B. Lennox, of Hammersmith Hospital, for so kindly lending me a typescript of his article; and Mr. W. Smith, of the Photographic Department, King's College Hospital, for the photographs and microphotographs.

REFERENCES

EISENBERG, A. A., and WALLERSTEIN, H. (1932), *Arch. Path.*, **13**, 716.
EWING, J. (1940), *Neoplastic Diseases*, 4th ed. Philadelphia and London: Saunders.
LANGHANS, T. (1907), *Virchows Arch.*, **189**, 69.
LENNOX, B. (1948), *J. Path. Bact.*, **60**, 295.
MARTIN, J. D., and ELKIN, D. C. (1939), *Ann. Surg.*, **110**, 169.
MAXIMOV, A. A., and BLOOM, W. (1942), *Text Book of Histology*. Philadelphia: Saunders.
MEANS, J. H. (1937), *The Thyroid Gland and Its Diseases*. Philadelphia: Lippincott.
RAWSON, R. W., and others (1948), *West. J. Surg.*, **56**, 82.
REIMANN, D. L. (1943), *Bull. School Med. Univ. Maryland.*, **28**, 93.
SMITH, L. W., and others (1934), *Amer. J. Cancer*, **20**, 1.
WILENSKY, A. O., and KAUFMAN, P. A. (1938), *Surg. Gynec. Obstet.*, **66**, 1.

GIANT HYPERTROPHIC GASTRITIS

A SURVEY OF THE LITERATURE AND THE RECORD OF A CASE TREATED SURGICALLY

BY W. R. FORRESTER-WOOD, HOVE

GIANT hypertrophic gastritis, a disease characterized by marked enlargement of the gastric folds, is rare and little is known of its aetiology, pathology, and treatment. Its recognition is important, for unless its distinctive features are elucidated a mistaken diagnosis of malignant disease may be made.

Massive enlargement of the gastric folds may be found either in localized areas of the stomach—and when this is so it may be associated with a previous gastric operation—or involving the whole of that viscus. In the latter cases it has usually been impossible to discover any predisposing cause.

Frequency.—The frequency with which giant hypertrophic gastritis occurs is very low and only about 40 cases have been recorded in the literature. Maimon, Bartlett, Humphreys, and Palmer (1947) report 6 surgically verified cases in 5765 gastroscopic examinations, together with 4 other cases diagnosed on the basis of X-ray and gastroscopic findings, giving an incidence of 0·17 per cent. These authors have reviewed the literature and have found 1 case recorded by Balfour (1919) in 8000 autopsies; 2 cases reported by Carmen in a series of 50,000 X-ray examinations; 1 case by Eliason and Wright (1925) in 8000 autopsies; 2 cases reported by Strauss, Meyer, and Bloom (1928), who had found 7 others in the literature up to that date; 3 cases in 2500 radiological investigations by Kantor (1936); 1 case in 25,000 X-ray examinations recognized by Feldman (1943); and 3 observed gastroscopically by Schindler (1939). Spriggs and Marxer (1943) have reported 9 cases of polypoid gastritis in a series of 4424 X-ray investigations, in only 3 of which was the lesion widespread throughout the stomach.

Aetiology.—At present the cause of this disease is unknown although several hypotheses have been suggested. It has been thought that the effects of chemical, thermal, or mechanical irritants may predispose to the changes found or that infections may be the cause.

The occurrence of hypertrophic changes as a sequel to operations on the stomach such as gastroenterostomy or partial gastrectomy suggest a mechanical or chemical cause, as do the cases described by Spriggs occurring after excess of alcohol. An infective origin is put forward in another of Spriggs' series in which a generalized hypertrophic gastritis was found in the stomach of a lunatic who habitually swallowed rubbish.

Symptoms and Signs.—One of the chief difficulties in diagnosis lies in the lack of any definite symptomatology. In most cases a history can be obtained of some epigastric or upper abdominal discomfort, but this may bear no constant relation to the taking of food; ingestion may relieve the discomfort in some, aggravate it in others. Anorexia, nausea, eructation, and vomiting may occur. General symptoms may include loss of weight, weakness, lethargy, headaches, constipation, or diarrhœa. Anæmia and its effects may cause the patient to seek advice. Physical signs are conspicuously absent. A gastric tumour is not found and only rarely can any tenderness or unusual discomfort be elicited by palpation of the upper abdomen. Evidence of weight loss or anæmia may be found, but is far from constant.

Investigations.—The diagnosis of giant hypertrophic gastritis is usually made radiologically, by gastroscopic inspection, or at autopsy.

X-ray Findings.—Although characteristic appearances are produced by the enlarged gastric folds, it is not always easy to differentiate hypertrophic gastritis from diffuse polyposis or adenomatosis of the stomach, or, in those cases where the disease is localized, from carcinoma. The principal distinguishing feature is the presence of rounded filling defects protruding into the lumen of the stomach, particularly along the greater curvature, and it is found impossible completely to obliterate these defects by pressure. The mucosal pattern is difficult to visualize, and it may require much time and patience to demonstrate the deep sulci between the hypertrophic folds, particularly when an excess of tenacious mucus covers the hyperplastic areas.

When the intervening fissures are not rendered visible the appearances may suggest some extrinsic pressure on the stomach, but when adequately filled a fairly typical appearance may be obtained. Multiple polyposis or adenomatosis may at first be difficult to exclude, but in this disease multiple punched-out crater-like filling defects are seen and give the appearance of vacuoles enclosed within the curves of the stomach walls.

Gastroscopic Findings.—The classic studies on the normal gastric mucosa of Wolf and Wolff (1944) have drawn attention to the very wide variations that may be found in the appearances of the stomach and have led to a realization that too much reliance should not be placed on the findings of a single gastroscopic examination. But where changes in the mucosa are gross this criticism becomes less valid and when correlated with other findings it is improbable that the marked alterations of hypertrophic gastritis can be mistaken for the lesser changes that occur in the emotional states. Many writers have given accurate and detailed descriptions of the changes observed in the mucosa of the stomach, but in most cases there has been no accurate differentiation between chronic hypertrophic gastritis and true giant hypertrophic gastritis. This may be due to the extreme rarity of the latter condition or to a failure to appreciate the possible differences in the natural history of the two conditions. Chronic hypertrophic gastritis is probably a temporary phase of a disease process which may progress to an atrophic gastritis and both hypertrophic and atrophic changes are usually found coexisting. Giant hypertrophic gastritis, on the other hand, does not seem to be associated at any time with atrophic changes, nor is it usual to find erosions on the summit of the gastric folds.

The typical appearances of giant hypertrophic gastritis reveal the mucosa of the stomach thrown up into enormous tortuous smooth folds separated by deep narrow fissures. It is impossible to obliterate the folds by inflation, particularly along the greater curvature of the stomach. Tenacious clear mucus may be present covering the mucosa. In many cases the size of the folds may raise the suspicion of true polypi, but, as Schindler points out, these polyp-like formations are loose and spongy in appearance, slightly bluish in colour, and are almost transparent, whereas true polypi are solid, orange-red in colour, and completely opaque.

Pathology.—

Morbid Anatomy.—The stomach is considerably enlarged but its serous surface is smooth. There may be considerable enlargement of the blood-vessels, and the whole organ may appear pinker than normal. There is no obvious hardening or stiffening of the stomach, but palpation gives the impression of a 'bag of worms'. The muscular coats may be thickened, but the main changes are to be found in the mucosal lining. These consist of an enormous hypertrophy of the normal gastric folds, which become raised and tortuous and present a marked similarity to the cerebral convolutions—a likeness first noted by Menetrier (1888). A height of 3 cm. may be attained by these folds, which become so crowded together that the intervening spaces are but deep narrow clefts. The whole mucosa is markedly hyperæmic and bright red or even bluish in colour. Often there is an excess of clear mucus covering the whole of the innermost coat of the stomach. The mucosa remains freely mobile on the underlying submucosa.

The extent of these changes is variable, as it may involve the whole of the stomach from cardiac to pyloric openings. On the other hand, the antrum may escape or the changes can be localized to the mid-portion of the stomach. Finally the hypertrophic mucosa may be limited to an area immediately surrounding the stoma of a gastro-enterostomy or the junction of stomach and jejunum after a partial gastrectomy.

Œdema, thickening, and increased vascularization may be present in the submucous layers. These changes differ considerably from the thickened, firm, nodular changes of chronic follicular gastritis—the *état mammelonné*—and from the multiple warty projections of the diffuse polyposis or adenomatosis of the stomach.

Histology.—The characteristic changes of importance are to be found in the mucosal and submucosal layers of the stomach, and these may vary from case to case or from one area of the stomach to another area of the same stomach. Although in some cases there are no gross changes in the epithelial layers, in others a varying degree of metaplastic changes may be found. This metaplasia consists of a change in type of cell towards the colonic type of epithelium, so that columnar cells and goblet cells replace to a greater or lesser extent the acid-secreting and pepsinogenic cells of the normal mucosa. Indeed, so enormous may be the increase in the number of mucus-secreting cells that the glands may become large mucous crypts.

Associated with the degree of metaplasia of the surface epithelium is the extent of acid production, and it is possible to gauge with some accuracy the degree of change towards the colonic type of mucosa by an estimation of the free hydrochloric acid in the gastric juice. Thus a normal or near normal free-acid index is indicative of little metaplasia, and the lower the acid index the greater the extent of approximation of the epithelium towards the colonic type. In most recorded cases there has been some diminution of the free hydrochloric acid, but a complete achlorhydria is not a feature of giant hypertrophic

gastritis, in contradistinction to multiple polyposis of the stomach where absence of free hydrochloric acid is a constant finding (Sinclair, 1938).

The submucous layer of the stomach is filled with an interstitial exudate which may contain a large number of eosinophils. Mononuclear cells and neutrophils may sometimes be found, and in some cases plasma cells predominate.

Treatment.—The treatment of giant hypertrophic gastritis may be medical or surgical, and the choice must depend on the severity of the symptoms produced and upon a consideration of the ultimate prognosis.

Spriggs (Spriggs and Marxer, 1943) has shown that mild symptoms can be relieved by effective medical treatment and that the gross swellings observed gastroscopically and radiologically can disappear, but he finds that both symptoms and swelling tend to return if the régime advised is forsaken.

The salient features of treatment are the elimination of septic foci—particularly in the mouth and upper respiratory tract—and suitable diet. Such a diet resembles the plain mixed diets used after peptic ulceration, with milk and milky foods. Gastric lavage is thought to be the most valuable physical measure and usually consists of a pint of warm water containing a teaspoonful of sodium bicarbonate to be drunk an hour before a meal. This is the so-called ' self-lavage '. Normal saline or weak hydrogen peroxide may be used instead of the bicarbonate. A mild antiseptic such as sulphocarbolate of soda is of value. Surgical treatment aims at a complete removal of the affected area of the stomach and consists of a partial or complete gastrectomy. Such resections are undoubtedly justified in cases with progressive symptoms, in those which fail to respond to medical measures, and in the generalized and extensive lesions which involve the whole of the stomach.

The most satisfactory approach for the operation of total gastrectomy is a combined abdominothoracic one and the technique of Garlock (1947) gives a surgical exposure of the whole operative field that cannot be improved. It ensures an easy œsophagojejunal anastomosis and does away with the short jejunal loop with its attendant troublesome puddling in the proximal loop. A diagrammatic sketch of the completed operation demonstrates the method of obtaining a long jejunal stump for junction with the œsophagus, and the use of the ' Y ' anastomosis of Roux.

CASE REPORT

W. D., aged 27, a lorry-driver, was referred to the Royal Sussex County Hospital with a history of loss of weight, progressive œdema of the ankles, shortness of breath, and occasional dyspeptic symptoms of a mild nature. His weight-loss had commenced about eighteen months prior to admission, and shortness of breath, œdema of the ankles, and pallor had been noticed for three months, as well as lassitude and occasional palpitations. The digestive symptoms were vague and extended back about five months, when there had been a short bout of epigastric pain an hour after food associated with nausea but no vomiting. Latterly his appetite had been poor and he suffered from " indigestion ". There was no history of hæmatemesis or malæna. He had suffered from malaria in 1943. Whilst in the Middle East he had also had several mild attacks of diarrhœa. His habits were moderate—he smoked about twenty cigarettes a day and only took a little alcohol—although he was employed by a firm of brewers. His mother had died at the age of 44, but his father and two sisters were alive and well and none had suffered from any gastric disturbances.

On Examination.—A well-developed young man, over 6 ft. in height. He was pale but not jaundiced. The tongue was clean. There was considerable pitting œdema of the lower extremities and over the sacrum.

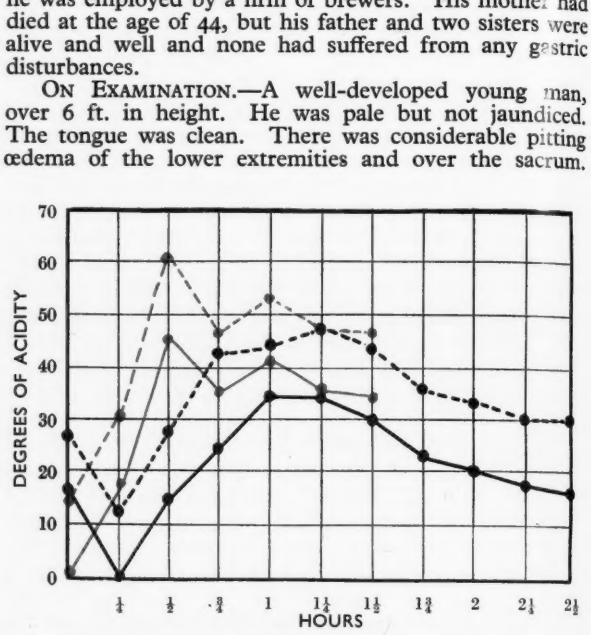

Fig. 371.—Gastric acidity curve. Test values in red, normal values in black.
●-----●-----Total Acidity; ●——●——Free HCl.

The heart was normal and blood-pressure 120/70. A few rhonchi and moist râles were audible at the right base.

Abdominal examination elicited no tumour or tenderness and, apart from a right lower abdominal paramedian scar, no abnormality could be detected. Rectal examination was negative, the C.N.S. showed no dysfunction, and urine analysis was normal. The blood-count showed a marked hypochromic anæmia, with a total red-cell count of 2,900,000 cells per c.mm. and Hb of 26 per cent (Haldane). The W.B.C. numbered 10,300 per c.mm., with 43 per cent lymphocytes. Fragility, bleeding, and clotting times and platelet count were normal. Van den Bergh direct reaction was negative and the blood W.R. negative. No occult blood (one examination only) was found in the stools but *E. histolytica* was present in vegetative and encysted forms. The histamine test-meal showed a normal curve (*Fig.* 371). X-ray examination of the chest was normal.

A barium meal was given and the radiologist reported that there were extensive filling defects affecting the greater curvature of the stomach from the proximal portion of the antrum to the fundus (*Fig.* 372). A month later a further report stated that the stomach again showed extensive filling defects affecting the greater curvature of the stomach and numerous projections into the gastric lumen (*Fig.* 373). It was considered probable that the changes indicated a benign condition.

Gastroscopy was performed by Dr. W. A. Bourne and showed an irritable stomach resistant to inflation. On the greater curvature and anterior wall nodular protuberances covered by reddened mucosa could be seen. The pylorus and antrum were not seen.

During the time these investigations were being carried out the patient's anæmia was treated and he was given a course of emetine for his amœbiasis.

At this point I was asked to see him with a view to surgical treatment—a provisional diagnosis of a diffuse benign lesion of the stomach having been made—and a gastric resection was suggested.

AT OPERATION.—On Aug. 11, 1947, I explored the abdomen through a left pararectal upper abdominal incision. The stomach was considerably enlarged and although the serous coat appeared normal it was evident from palpation that the inner coats were the seat of widespread changes as the organ felt like an enormous varicocele and was obviously involved from the pylorus to the cardia. The incision was therefore extended across the costal cartilages and upwards and outwards through the seventh intercostal space. The suspensory ligament of the left lobe was divided and the diaphragm split down to the œsophageal hiatus, after crushing the left phrenic nerve as it coursed over the pericardium. A total gastrectomy was performed, a retrocolic œsophago-jejunal anastomosis, and a Roux jejunojejunal anastomosis 'en Y' effected after the technique of Garlock (1947).

A valvular jejunostomy below the jejunojejunostomy was made; the left pleural space was drained through a stab-wound and the main incision closed in layers. (*Fig.* 374.)

A transfusion was given during the operation and carried through to the post-operative phase and oxygen through a B.L.B. mask was started at the conclusion of the operation. The anæsthetic consisted of pentothal, cyclopropane, and curare.

PROGRESS.—The post-operative convalescence was relatively smooth. No fluids were swallowed for four days and the jejunostomy tube was removed on the sixteenth day. At the end of three weeks he was taking a light diet well and was discharged from hospital 3½ weeks after operation.

One month later he was shown at the Clinical Section of the Royal Society of Medicine and was then taking four normal meals a day without discomfort. Two months after leaving hospital he reported having gained over a stone in weight and he was completely symptom-free.

PATHOLOGICAL REPORT.—The stomach was enlarged and felt like a bag of worms. The serous coat was smooth and the vessels were large and prominent. The organ was opened along the greater curvature, revealing an area of mucosa approximately 35 cm. × 25 cm. in size. The mucosa was thrown into bluish-red hypertrophied folds varying from 0·5 cm. to 2 cm. in height.

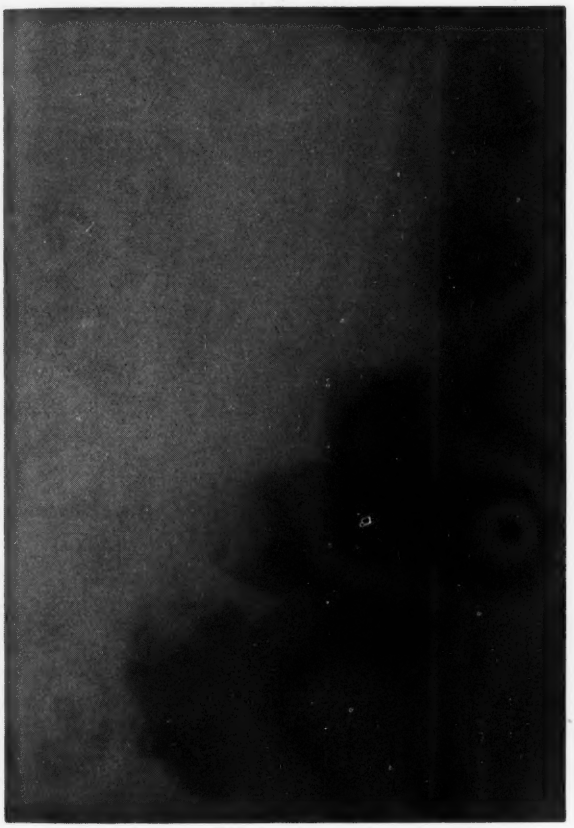

FIG. 372.—Radiograph showing large smooth filling defects extending along the greater curvature of the stomach.

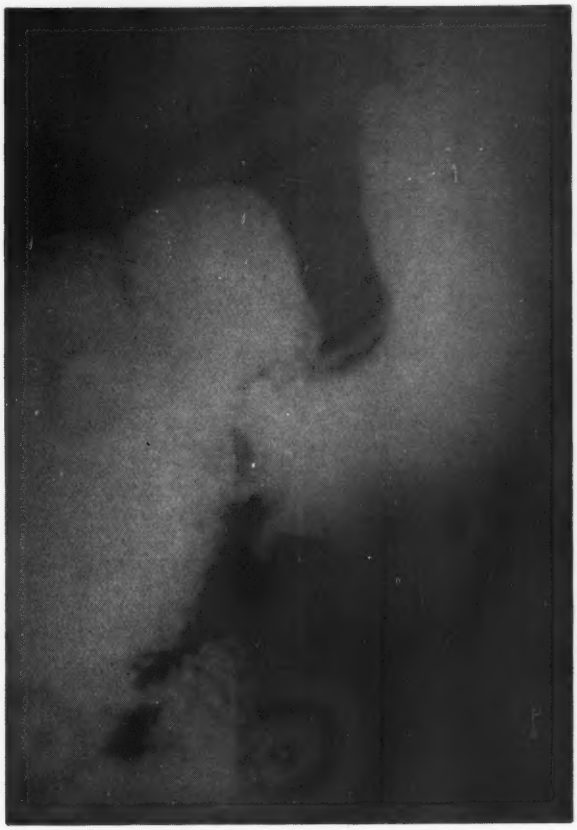

FIG. 373.—Radiograph showing large filling defects along greater curvature of the stomach extending right up to the pyloric antrum.

The resemblance to cerebral convolutions suggested itself at once (*Fig.* 375). These enormously hypertrophied mucosal folds showed a diffuse mammilation, moved freely over the muscle coats, and were present throughout all parts of the stomach from cardia to pylorus.

Microscopical examination showed that the large folds of mucosa were largely formed by hyperplastic glands in which the epithelial elements were present in relatively normal proportions. There was little sign of intestinal metaplasia. The interstitial tissue between the glands showed some œdema and numerous cells, chiefly eosinophils and plasma cells. An occasional gland, usually one near the muscularis mucosæ, was cystically dilated. The submucosa was thickened by an increase in collagenous tissue and in some areas by œdema. Relatively dense cellular infiltration, largely perivascular in position, of lymphocytes and plasma-cells, with some eosinophils, was also present. Fine cores of collagen and of the muscularis mucosæ, together with plasma and eosinophil cells, extended up in support of the hypertrophied mucosal folds. The muscle coats showed some hypertrophy in the body of the stomach but elsewhere appeared of normal width. The cellular

exudate seen in the submucosa also extended perivascularly into the muscle coats in some areas. The serosa was normal. There is nothing to suggest malignant change.

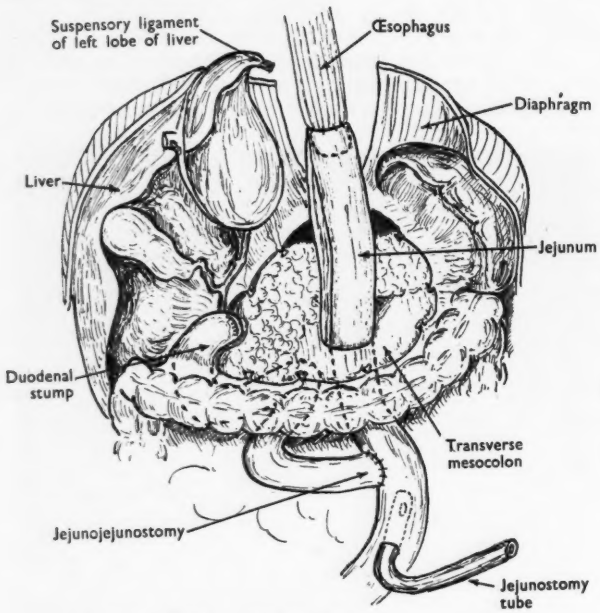

FIG. 374.—Sketch showing final stage of operation. (After Garlock.)

SUMMARY AND DISCUSSION

A case of generalized hypertrophic gastritis is presented, with a review of the literature and a summary of the salient features of frequency, aetiology, diagnosis, pathology, and treatment.

The case serves to illustrate the severity of the symptoms that may arise, the diagnostic investigations necessary to reach a correct diagnosis, and the details of the operation of total gastrectomy which may have to be performed in some cases. No further light has been shed on the aetiological factors, although it is worth noting the association with amœbiasis. Previously recorded cases have been known to occur in association with ' dysentery '. It is unfortunate that no estimation of the plasma-protein was made in this case, since Maimon and others (1947) have pointed out the possible role of hypoproteinæmia. Dietary insufficiency is known to be associated in some cases with œdema of the gastric mucosa, and in the case here recorded peripheral œdema was a marked feature when the patient first presented.

The possible relationship between giant hypertrophic gastritis and malignancy has been discussed in papers already published, and it seems fairly clear that despite the metaplastic changes that may be found in gastric mucosa there is no evidence to support the hypothesis that it is a pre-cancerous condition.

FIG. 375.—Photograph of stomach after being opened along the greater curvature, showing gross hypertrophy of mucosa.

I wish to record my acknowledgements to Dr. R. Kemball Price for referring the case to me; to Dr. W. A. Bourne for the gastroscopic findings; and to Dr. L. R. Janes for the pathological report. I am also indebted to Dr. Bourne and Dr. Janes for their help and criticism in the writing of this paper.

REFERENCES

BALFOUR, D. C. (1919), *Surg. Gynec. Obstet.*, **28**, 465.
ELIASON, E. L., and WRIGHT, V. M. W. (1925), *Ibid.*, **41**, 461.
FELDMAN, M. (1943), *Radiology*, **41**, 181.
GARLOCK, J. H. (1947), *J. thorac. Surg.*, **16**, 215.
KANTOR, J. L. (1936), *Amer. J. Roentgenol.*, **35**, 204.
MAIMON, S. N., BARTLETT, J. P., HUMPHREYS, E. M., and PALMER, W. L. (1947), *Gastroenterology*, **8**, 397.
MENETRIER (1888), quoted by SPRIGGS and MARXER.
SCHINDLER, R. (1939), *Amer. J. digest. Dis.*, **6**, 523.
SINCLAIR, N. (1938), *Brit. J. Surg.*, **20**, 645.
SPRIGGS, E. I., and MARXER, O. A. (1943), *Quart. J. Med.*, **12**, 1.
STRAUSS, A. A., MEYER, J., and BLOOM, A. (1928), *Amer. J. med. Sci.*, **176**, 681.
WOLF, S., and WOLFF, H. G. (1944), *Human Gastric Function*. Oxford: University Press.

WILMS' TUMOUR

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THE fact that these tumours are relatively uncommon and that the survival of proved cases for more than five years is extremely rare, justifies the report of this case in which the patient is still alive and well six years and eight months after the original operation.

CASE REPORT

R. E. M. C., born June 3, 1937, was admitted to hospital on April 18, 1942, with a history of diarrhœa and vomiting for the past 24 hours. He had vomited six times and the vomit was greenish in colour. His mother stated that the child had never had any previous illness and that no other member of the family was affected by similar symptoms. The mother further stated that she had noticed a lump in the right side of the abdomen one week previously, but she did not worry about it because the child did not complain of any pain or discomfort.

ON EXAMINATION.—The temperature, pulse, and respiration were 97°, 104, and 26 respectively. He was a perfectly healthy looking child with a slightly flushed face. The tongue was clean, but dry, and the fauces showed no abnormality. The heart and lungs were examined but again no abnormality was detected. On examination of the abdomen, however, a large mass was visible and palpable in the right hypochondriac region and right loin and it was estimated to be about the size of a coco-nut. The surface was slightly irregular and it was of a firm consistence and felt solid. There was no pain, tenderness, or muscular rigidity; the mass was very mobile and could be pushed easily into the loin. It could also be separated from the liver, with which it did not appear to have any connexion. The liver and spleen were not palpable, nor was the left kidney. The percussion note was resonant anteriorly, but dull posteriorly. The bladder was not distended and no abnormality was detected on rectal examination. A provisional diagnosis of Wilms' tumour was made; further investigations helped to confirm this.

A report on a mid-stream specimen of urine was as follows: Acid; a few epithelial cells and calcium oxalate crystals. Culture, sterile.

Other reports were :—

1. Blood-urea: 20 mg. per cent.
2. Urea-concentration test :—

Specimen	*Volume*	*Urea*
0	90 c.c.	5·0 mg. per cent
1	38 c.c.	4·06 mg. per cent
2	60 c.c.	4·0 mg. per cent

3. Radiograph of chest: no abnormality detected.
4. Straight X-ray of renal tract: right kidney outline cannot be seen apart from outline of the tumour.
5. Intravenous pyelogram: right kidney displaced upwards and medially. Slight distortion of lower calix. Left kidney normal.

Fig. 376.—Wilms' tumour of the right kidney from a boy aged 4½ years, showing the smooth capsule of the tumour.

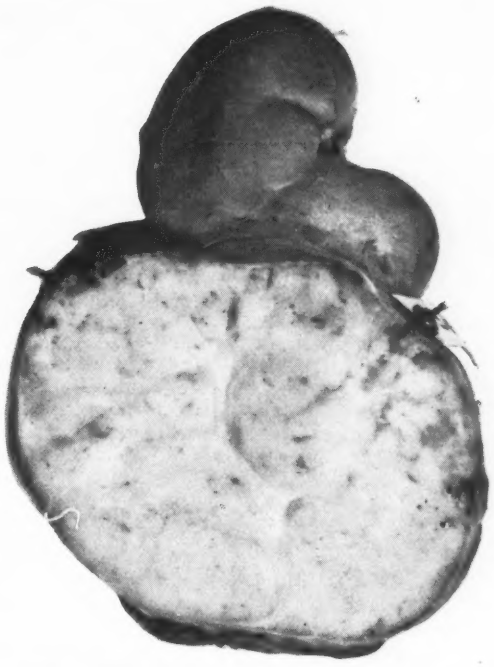

Fig. 377.—Showing the cut surface of the tumour.

6. Hæmoglobin, 90 per cent.
7. Blood-count :—

White blood-corpuscles	8125	
Polymorphonuclear leucocytes	52	per cent
Eosinophils	0	,, ,,
Lymphocytes	37	,, ,,
Monocytes	11	,, ,,

8. Blood Group B. Father, Group B; Mother, Group O.

AT OPERATION.—The patient was given atropine sulphate gr. $\frac{1}{100}$ and seconal gr. ½ per 10 lb. of body-weight half an hour before operation (child's weight, 2 st. 5 lb.). Gas, oxygen, and ether were administered at the

operation, during which a transperitoneal nephrectomy was performed. The kidney area was drained by a rubber drain inserted through a stab incision in the loin. Two pints of blood were given both during and after the operation, followed by intravenous saline and 5 per cent glucose.

PROGRESS.—Convalescence was entirely satisfactory, the stitches being removed on the thirteenth day, and he was discharged on May 15.

Post-operative deep X-ray therapy was started on June 10, and it was completed on June 26. A total of 2250 r was given in equal doses to the anterior, posterior,

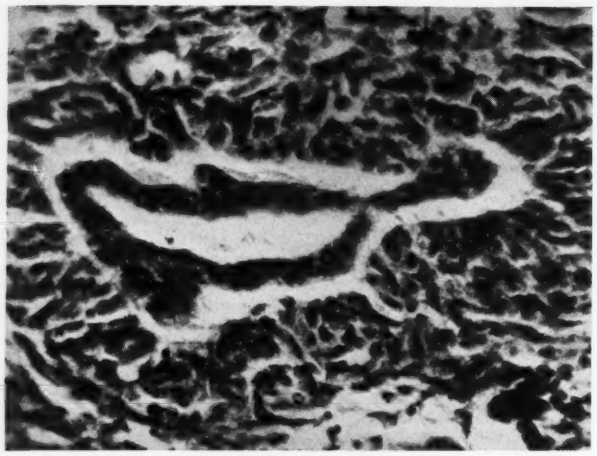

Fig. 378.—Microphotograph of tumour showing an irregularly shaped tubule lying in a cellular stroma. (× 150.)

and lateral abdominal areas, and no further irradiation has been given.

The patient, now 11 years old, has remained exceptionally well since his operation apart from a mild attack of measles. His height is 4 ft. 8 in., weight 5 st. 2 lb., appetite excellent, and he has no dysuria or hæmaturia. He plays football, cycles, swims, does physical training, and altogether behaves like any normal child of his age. His school reports are also excellent.

PATHOLOGICAL REPORT (*Figs.* 376–378).—A firm round encapsulated tumour in the lower pole of the right kidney. Diameter of tumour 4 in. Microscopical appearance: picture is that seen in a Wilms' tumour.

DISCUSSION

PATHOLOGY

Campbell (1937) studied a series of cases of primary malignant tumours of the kidney in infants and children and reported that the vast majority were Wilms' tumours and that there were only 2 cases of carcinoma and 2–11 per cent hypernephromata. He also observed that tumours of the renal pelvis and fibrous capsule were rare. The following analysis of three further series of cases is also interesting :—

Author(s)	*No. of Cases of Primary Tumour of Kidney*	*No. of Wilms' Tumours*
1. Smith and Young (1941)	118	2
2. Deeming (1946)	82	6
3. Harvey (1947)	54	3

Nowlin (1940) states that the familial tendency to renal malignancy appears to be unusual.

In malignant renal tumours it is usually stated that the female is affected twice as often as the male, but in Wilms' tumour the sexes appear to be equally involved. In considering age, it must first of all be pointed out that these tumours may be congenital or acquired within the first few years of life. The youngest patient I have been able to trace in the literature is that described by Levinson and Furst (1937) in a child three weeks old.

There seems little doubt that the majority of cases occur between the first and third year of life and are relatively uncommon after 5 years; they may, however, be found in adults. Weisel, Docherty, and Priestley (1943 a, b) reported on 44 cases in which 36 cases were under 9 years of age and the others between the ages of 12 and 59 years. Nesbitt and Adams (1946) reported on another series of 16 cases in which the youngest case diagnosed was 6 months old and where 15 of the cases were under 6 years of age. The vast majority of the tumours occur in young subjects, but Eserky and others (1947) have described 3 cases occurring in adults and state that altogether 57 adult cases have been reported. Hill (1946) also reported on a case in a female aged 45.

The origin of Wilms' tumour is still a matter of controversy, but the following are the theories which are mainly accepted. It originates: (1) As an embryonic cellular rest; (2) As a teratomatous development of one or more germ layers; (3) In the region of the metanephros proper; (4) As a perverted growth of the renal mesenchyme.

The tumour may be found in any of the renal parenchyma and is nearly always unilateral, although a few bilateral cases have been reported. Stern and Newns (1938) came to the opinion that the tumour was commoner on the left side. The majority of the tumours are found in the lower pole and are of a soft consistence, with a smooth or nodular surface. McGinn and Wickham (1946) reported on a case in a female child 2 years of age which occurred in a horseshoe kidney. The tumour is usually described as being encapsulated by a tough fibrous capsule with the renal tissue displaced and distorted by pressure. Late in the disease the capsule is perforated and the kidney tissue is involved. According to Vuori (1947) the capsule consists of a layer of renal tissue which has sclerosed into connective tissue due to the pressure of the slowly-growing tumour, but in rapidly-growing tumours the capsule is imperfect. If the tumour perforates the capsule, it invades the perirenal cellular tissues and becomes adherent to the posterior abdominal wall and the adjacent organs. It may spread along the renal vein and give rise to a tumour thrombosis. It may even extend into the inferior vena cava and the spermatic vein, completely or partly obliterating the lumen. Emboli may thus be carried to produce distant secondaries. The occurrence of these secondaries has been variously estimated at 30–40 per cent of all the cases seen, but metastases in the regional lymph-nodes are only present in very advanced cases. Local recurrence may be found fairly frequently after removal and the incidence of secondaries is given as :—

Lungs 44 per cent Liver 20 per cent	Lymphatic glands Bones	} 8 per cent

The commonest site in bones is the upper end of the humerus or femur and may result in pathological fracture. According to Stern and Newns (1938) vesical metastases are very uncommon.

If the tumour is sectioned after nephrectomy and examined with the naked eye, it will be noticed that the surface glistens and that it has a much more uniform appearance than the Grawitz's tumour. It varies from a white to a brownish colour and there are areas of hæmorrhage or necrosis; small cystic areas may also be present. It is true to say that, in children, only microscopical examinations will reveal the true nature of the tumour.

Microscopical Appearance.—It is noticed that the blood-supply is very poor and that the vessels have very thin walls. The structures observed are extremely diverse. Cartilage, bone, and striated and plain muscle have all been described, along with round or spindle-shaped cells of a sarcomatous nature lying in a scanty delicate matrix. Acinar or columnar and cubical adenomatous cells are also arranged irregularly.

CLINICAL PICTURE

I think it is perfectly true to say, as in my own case, that there are usually no symptoms in the early stages and that the tumour is first noticed by the parents because of the increase in the size of the abdomen. In some cases there may be a rapidly-growing abdominal mass, but this is not the usual history. If the diagnosis has been missed and the child is brought to the surgeon in the later stages there is a history of rapidly deteriorating health, loss of weight, pallor, gastro-intestinal disturbances, and abdominal pain. The doctor or mother may also state that the child is running a temperature and that this is the only symptom. If a mass is palpable in the loin and there is no clinical evidence of renal infection, then Wilms' tumour should be suspected. This point was stressed by McCague (1940).

It is not my intention to give in detail the clinical examination of renal tumours, but the following points are most important:—

1. Do not conclude that because the kidney is enlarged that it is necessarily pathological, because it may be due to a compensatory hypertrophy.
2. The mass lies in the loin, or can be pushed into the loin.
3. The mass is more or less kidney-shaped.
4. The mass moves with respiration, except in late cases where it is adherent to the surrounding structures.
5. There is a band of resonance anteriorly due to gas in the ascending or descending colon.
6. There is dullness on percussion posteriorly.
7. Recent and rapid development of a varicocele.
8. Tumours of the kidney grow downwards and forwards in contradistinction to a perinephric abscess, which enlarges posteriorly.
9. If the tumour is very large, the following additional points should be noted:—

a. The lower ribs are displaced outwards;

b. The diaphragm is displaced upwards, thus producing dyspnœa.

c. Ascites appears, due to pressure on the inferior vena cava.

10. Proteinuria is usually present and is thought to be due to congestion of the kidney.

Swelling.—Rowe and Frazer (1944) pointed out that rapid growth is more prevalent than hæmaturia and pain. This swelling, which is occasionally bilateral, is usually described as a firm reniform mass, often nodular, and in the later stages becomes quite fixed.

If there is any doubt about the mobility of the tumour, it can be determined by radiological examination with the patient first in the lying and then in the upright position. It is also convenient to mention here that aspiration biopsy has no place in the diagnosis of renal tumour.

Hæmaturia.—In cases of Wilms' tumour there may be no history of hæmaturia when the child is brought for examination, or if there is, it is usually a late symptom. It has been estimated that it occurs in 15–20 per cent of all cases.

This bleeding may occur from: (1) Invasion of the pelvi-calical system; or (2) Congestion of the kidney, but is rarely severe. Needless to say, it is absolutely essential that all cases complaining of this symptom must be thoroughly investigated to determine the true cause. The investigations must include routine medical examination, complete blood-count, examination of catheter specimen of urine, straight X-ray of renal tract, cystoscopy and pyelography, etc.

Pain.—Pain is usually preceded by hæmaturia and is usually described as a dull, inconstant ache in the loin or back. The pain, however, may be very severe and colicky, and in these cases it is usually due to the passage of blood-clots down the ureter. The dull aching pain is usually attributed to one or more of the following causes:—

1. Increased pressure within the renal capsule due to: (*a*) Slow or rapid growth of tumour; (*b*) Hæmorrhage into the tumour.
2. Obstruction to the outflow of urine at the pelvi-ureteric junction.
3. Pressure or traction on the renal pedicle.
4. Involvement of nerves.
5. Secondaries, causing: (*a*) Backache; (*b*) Referred pain along the various parietal nerves, e.g., sciatica; (*c*) Headache; (*d*) Bone pain according to situation of secondary growth; (*e*) Visceral discomfort.

It has been estimated, however, that the renal tumour triad (hæmaturia, pain, and abdominal tumour) occurs only in approximately 20 per cent of cases.

Cystoscopy and Pyelography.—It is hardly necessary to mention that the age of the patient may prevent cystoscopy and retrograde pyelography. If intravenous pyelography is impossible, then it is still possible to obtain satisfactory X-ray films by giving an intramuscular injection of 35 per cent solution of diotrast. (Rowe and Frazer, 1944.) Cystoscopy should be carried out for the following reasons: (1) To exclude a vesical lesion; (2) To demonstrate blood issuing from the ureteric orifice; (3) To test the functional capacity of the presumably normal kidney.

It is generally agreed now that retrograde pyelography is better than intravenous pyelography for all doubtful cases, and especially in the early stages.

Whichever method is used, however, the following alterations from a normal pyelogram may be observed: (1) Kidney may be enlarged; (2) Displacement of kidney; (3) Kidney pelvis distorted, compressed, or dilated; (4) Failure to visualize renal pelvis or filling defect present; (5) Hydronephrosis; (6) Elongation or distortion of calix or calices; (7) Complete or partial amputation of one or more calices; (8) Ureter takes an abnormal course and may be bowed at the upper end.

In cases of congenital polycystic disease of the kidneys, the calices are shortened and broadened, whilst in retroperitoneal tumours there is displacement of the kidney and ureter without distortion of the renal pelvis.

DIFFERENTIAL DIAGNOSIS

Before discussing this section it is wise to point out that perirenal insufflation is often dangerous and that this investigation is really of very little value. It is obviously impossible in an article of this type to go into all the details of a differential diagnosis, but the salient points will be stressed.

1. Kidney Swellings.—

a. Congenital polycystic disease of the kidney—usually bilateral and nodular. Hæmaturia common.

b. Congenital abnormality of kidney, e.g., unilateral fusion and congenital hydronephrosis.

c. Large ' retention ' cyst, associated with chronic interstitial nephritis.

d. Hydatid cyst of kidney—invariably in adults.

e. ' Serous ' cysts of kidney—invariably in adults.

f. Hydronephrosis and hæmorrhage into it—obstruction in some part of urinary tract, e.g., stricture, calculus, aberrant renal arteries, etc.

g. Tuberculous hydronephrosis—other signs of tuberculosis.

h. Hypernephroma—usual age 30–50.

i. Sarcoma of kidney—very rare and only diagnosed on microscopical examination.

j. Simple tumours of kidney, e.g., lipoma, fibroma, angioma, and adenoma. Usually small, pale yellow, and encapsulated. Rarely attain very large size.

k. Carbuncle of kidney—history of rigors, sweating, pain in loin, and emaciation.

l. Pyelonephritis—previous history of pyelitis, bladder operation, etc.

m. Pyonephrosis—history of infection of kidney and renal pelvis.

n. Perinephric abscess—previous renal history usual.

2. Adrenal Tumours.—

a. Cortical: (i) Diffuse hyperplasia—may be due to disturbances of cholesterol metabolism. (ii) Adenoma—virilism in male child or " Infant Hercules " type. Virilism in female child. Excessive production of androsterone. (iii) Carcinoma—similar to hypernephromata.

b. Medullary: (i) Pheochromocytoma — rare. Marked rise in blood-pressure, etc., due to sympathetic stimulation. (ii) Neuroblastoma—highly malignant with early metastases. (iii) Ganglioneuroma—rare.

3. Sarcoma involving retroperitoneal tissues of bowel.

4. Pancreatic cyst.

5. Mesenteric cyst.

6. Leukæmic infiltration of liver and spleen.

7. Splenic enlargement, e.g., cysts, tumours, Banti's disease, etc.,

8. Hepatic enlargement, e.g., angiomata, cysts, etc.

9. Ovarian neoplasm.

10. Tuberculous peritonitis.

11. Sympathicoblastoma arising from autonomic system.

12. Retroperitoneal cysts, e.g., developmental:—

a. Lymphatic.

b. Enterogenous.

c. Urogenital.

d. Mesocolic.

PROGNOSIS

Briefly, the prognosis generally may be said to be very bad, and according to Harvey (1947) the survival period depends upon the size of the tumour, the age of the patient, and the degree of malignancy. Marked loss of weight, evening pyrexia, anæmia, and a fixed palpable tumour definitely denote a bad prognosis, and death usually results from general dissemination, exhaustion, or pulmonary embolism.

Wiesel, Docherty, and Priestley (1943) reported on 44 cases in which 31 died within 1 year of operation, 6 more within 4 years, and only 7 were living after periods of from 2 to 20 years.

Vuori (1947) gave his immediate operation mortality as 2 out of 18 cases and stated that 4 of his patients were still alive, 3 after 1 year, one of which already had secondaries.

Priestley and Schulte (1932) analysed 39 cases and found that only 6 were alive after 5 years or more. There is, unfortunately, no unanimity of opinion, but generally it can be stated that the prognosis is more hopeful if radiological treatment is given pre-operatively and/or post-operatively.

TREATMENT

There is no doubt that nephrectomy is the only method of treatment which holds out any hope of a cure, and it is now invariably accompanied by pre-operative and/or post-operative deep X-ray therapy.

Radiological Treatment.—It is also generally agreed that radiological treatment alone will not cure the patient, but it would appear that it does reduce the incidence of metastases provided that the tumour is radiosensitive. Levinson and Furst (1947) reported on 2 cases in which one was radiosensitive and the other radio-resistant; the former was alive and well 9 years later, but the latter died within 3 months of operation. Vuori (1947) reported on 9 cases which were treated post-operatively with deep X-ray therapy, but all died.

According to Rowe and Frazer (1944), the advantages of pre-operative irradiation are: (1) Reduction of tumour minimizes operative manipulation, but according to Ladd there is no need to shrink the tumour. (2) Production of fibrosis diminishes vascularity. (3) Patient comes to operation in good condition. (4) Rapid response of these tumours to irradiation confirms the diagnosis.

Deeming (1946), however, advises against pre-operative X-ray therapy and states that irradiation of the lung area may produce fibrotic pneumonitis.

Another objection to pre-operative irradiation is the danger of distant metastases during the 3- to 6-week interval before operation, but Priestley and Shulte (1942) do not consider this argument to be a convincing one. Weisel, Docherty, and Priestley (1943) are agreed that if X-ray therapy is given pre-operatively, the sole aim should be to reduce the tumour to operable dimensions and the period of its application should not exceed three weeks. Nesbitt and Adams (1946) and Rowe and Frazer (1944) recommend nephrectomy followed by irradiation. Campbell gives his mortality as 95 per cent when treated by nephrectomy alone and advises pre-operative and post-operative deep X-ray therapy. Kaplan and Campbell (1939) reported on a male child of 11 months to whom irradiation was given daily (200 r per treatment) alternately to the anterior, posterior, and lateral abdominal areas until a total of 1200 r had been given to each area. The tumour was greatly reduced, and two months later a well-encapsulated tumour of the right kidney was removed. Post-operatively 800 r were given over the anterior and posterior abdominal areas and further courses have been given at intervals. The child was alive and well three and a half years later. Pre-operatively the patient should also receive a full diet, iron therapy, and, if necessary, a blood transfusion. For obstinate hæmaturia the renal pelvis may be lavaged with 1 per cent silver nitrate.

Operation.—Vuori (1947) reported on 18 cases in which the transperitoneal approach was used in 5 cases and the retroperitoneal route in 13 cases.

Sweetser (1947) suggests the following approach to the kidney in cases of large malignant tumours. He uses the routine lumbar incision and extends it obliquely across the abdomen to the middle line at a point midway between symphysis and umbilicus. It is then extended vertically upwards in the midline to the xiphoid. The large triangular flap is then lifted upwards to give an excellent exposure with a minimum of injury to the vessels and nerves of the abdominal wall.

The advantages of this incision and the transperitoneal approach are: (1) Allows exposure, clamping, and division of the vascular pedicle before the tumour is disturbed. (2) Prevents metastases by blood-stream by squeezing out tumour cells. (3) Gives excellent exposure of the tumour and enables operator to avoid important displaced structures, e.g., inferior vena cava.

Deeming (1946) also suggests that the inferior vena cava should be opened if there is a prolongation of tumour tissue into it. It is important to note that metastases are *not* a contra-indication to operation, as they are usually sensitive to radiotherapy.

SUMMARY

1. A case of Wilms' tumour alive and well six and a half years after operation.

2. Treatment was by nephrectomy and post-operative deep X-ray therapy.

3. Review of literature.

I wish to express my thanks to Mr. I. Lewis, Senior Surgeon, North Middlesex Hospital, London, N. 18, for permission to publish this case.

BIBLIOGRAPHY

CAMPBELL, M. F. (1937), *J. Amer. med. Ass.*, **109**, 1606.
DEEMING, C. L. (1946), *J. Urol.*, **55**, 571.
ESERSKY, G. L., ET AL. (1947), *Ibid.*, **58**, 397.
HARVEY, N. A. (1947), *J. Urol.*, **57**, 669.
HILL, R. M. (1946), *Brit. J. Urol.*, **18**, 53.
KALPAN, I. I., and CAMPBELL, M. F. (1939), *Arch. Pediat.*, **56**, 766.
LEVINSON, L. J., and FURST, N. J. (1947), *J. med. Soc. N.J.*, **44**, No. 1, 8.
MCCAGUE, E. J. (1940), *Arch. Surg., Chicago*, **41**, 385.
MCGINN, E. J., and WICKHAM, J. M. (1946), *J. Urol.*, **56**, 520.
NESBITT, R. M., and ADAMS, F. M. (1946), *J. Pediat.*, **29**, 295.
NOWLIN, P. (1940), *J. Urol.*, **43**, 654.
PRIESTLEY, J. T., and SCHULTE, T. L. (1942), *J. Urol.*, **47**, 7.
ROWE, E. W., and FRAZER, M. D. (1944), *Radiology*, **42**, 107.
SMITH, E., and YOUNG, A. (1941), *J. Urol.*, **45**, 519.
STERN, R. O., and NEWNS, G. H. (1938), *Arch. Dis. Childh.*, **13**, 193.
SWEETSER, T. H. (1947), *Ibid.*, **57**, 651.
VUORI, E. E. (1947), *Act. chir. Scand.*, **95**, 555.
WEISAL, W., DOCHERTY, M. B., and PRIESTLEY, J. T. (1943, a), *J. Urol.*, **50**, 399.
— — — — — — (1943, b), *Ibid.*, 564.

SOLITARY MYELOMA WITH GENERALIZED METABOLIC DISTURBANCE

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ALTHOUGH multiple myelomatosis is now a well-recognized clinical entity, solitary myeloma of bone characterized by a single tumour-mass showing no evidence of dissemination throughout its course, remains a rare disease. The literature was reviewed by Willis in 1941 when he accepted only 13 cases (including his own case) as true examples of the condition. Since then cases have been reported by Gootnick (1945), Kaufman (1945), and Lumb (1948), bringing the total number of recorded examples up to 16. There are, in addition, many recorded instances of patients presenting with apparently solitary myelomata which subsequently underwent generalization (Geschickter, 1930; Cutler, Buschke, and Cantril, 1936; Paul and Pohle, 1940; Gootnick, 1945). Many of the cases which these writers have included in their reviews are not, however, acceptable as true examples of solitary myeloma. In some generalization was seen to occur while in others the period of observation was too short to exclude that

possibility; and in many of the remainder repeated radiological examinations or full autopsy were not performed.

The frequency with which apparently solitary tumours have later become generalized raises the question as to whether the solitary forms are indeed always a prelude to the generalized disease. In the 16 cases referred to above, however, careful observation of the cases over a long period failed to reveal evidence of generalization, and Willis (1948) concludes that the solitary form is a distinct entity and not merely an early and localized stage of the general disease. The present paper records a further instance of a solitary myeloma of bone. The tumour had been present for at least five years before death and the absence of generalization was confirmed at autopsy. The special interest of this case lies in the occurrence of metabolic disturbances which are usually regarded as indicative of generalization, and in the diagnostic difficulties encountered.

Fig. 379.—Anteroposterior view of sacrum, January, 1948. There is cystic expansion of both wings, extending across the midline.

Fig. 380.—Radiograph of sacrum taken in 1943, showing already a well-developed cystic lesion.

CASE REPORT

H. B., an unmarried man aged 50 years, attended as an out-patient at St. George's Hospital in January, 1948, complaining of swelling of the feet, ankles, and legs of three months' duration. He also complained of widespread body pains commencing in 1940, following a fall from a bicycle. At first these pains were at the lower end of the spine, but later they spread down both legs. By the end of 1942 the pains had become much more severe and in March, 1943, the patient attended a London hospital. There he was X-rayed and a course of physiotherapy was given, but, as no improvement followed, spinal manipulations were later employed. These manœuvres only intensified his disability, which then remained essentially unchanged for three years. In 1946 the pains spread to his neck and down both arms to the hands and fingers. In January, 1947, a slipped cervical disk was suspected and cervical laminectomy was performed, but did not bring about any improvement, and in October, 1947, the patient developed the swelling of the lower limbs which led to his attendance at St. George's Hospital. Although his appetite was capricious he had not lost weight, and bowel function and micturition were normal. His earlier medical history and the family history contain nothing relevant to the case.

When examined at the out-patient department in January, 1948, he was a pasty-complexioned ill-looking man with rather pale mucous membranes. The tongue was clean and the fauces clear, and there was no glandular enlargement. The peripheral arteries and the optic disks were normal. The pulse was regular and the blood-pressure 120/75 mm. Hg. The heart was not enlarged, and the sounds were normal. Examination of the chest and of the abdomen were negative and there were no enlarged viscera on palpation. The nervous system likewise showed no abnormality, and rectal examination showed a normal prostate. Pitting œdema extended over the feet, ankles, and legs, as high as the knees, and was present also over the sacral region. There was also some puffiness of the face and hands. Examination of the urine showed a heavy cloud of protein, and arrangements were made to admit the patient to hospital for a full investigation.

SPECIAL INVESTIGATIONS.—Radiographs made in hospital showed the old laminectomy which had been performed at the level of the 6th cervical vertebra but the remainder of the cervical, dorsal, and lumbar spine appeared normal. The sacrum, however, showed an extensive destructive process with a cystic appearance (*Fig.* 379). Both wings of the sacrum were affected, the process extending across the midline anteriorly. Osteoclastoma, myeloma, and secondary renal carcinoma were considered as possible diagnoses and further radiographs of the entire skeleton were made. They showed,

however, no abnormality except for a slight degree of osteoporosis, and tomography of the sacrum added no further information. The hospital which the patient had attended in 1943 was then approached, and, although the old case records were not available, radiographs taken in 1943 were obtained, and these showed already a well-developed destructive and cystic lesion in the sacrum (*Fig.* 380).

Hæmatological examination showed a moderate degree of orthochromic anæmia (R.B.C. 3,600,000 per c.mm.; hæmoglobin 10 g. per 100 ml.) which persisted unchanged throughout the disease. The white-cell count remained around 10,000 per c.mm., with a normal differential count, except following radiotherapy when a transient eosinophilia (up to 21 per cent) occurred. Sternal marrow biopsy showed a normal cell distribution and no unusual cell forms were present. The erythrocyte sedimentation-rate (Wintrobe) was constantly elevated, giving values between 36 and 62 mm. in 1 hour. The Wassermann and Kahn tests were negative. Urine examinations showed a persistent moderate proteinuria (50–100 mg. per 100 ml.). The deposit contained a few hyaline casts and red blood-corpuscles. Preliminary chemical examination of the blood gave the following results: Urea, 42 mg. per 100 ml.; total serum-protein 5·2 per cent (albumin 2·8, globulin 2·4); alkaline phosphatase 8·3 units; acid phosphatase 2 units; serum-calcium (mean of three estimations) 12·2 mg. per 100 ml.; serum inorganic phosphorus (mean of three estimations) 4·2 mg. per 100 ml. In view of the persistently raised serum-calcium figures a metabolic balance experiment was carried out over a three-day period after preliminary stabilization on a low-calcium diet. The findings are recorded in the accompanying table. The urinary output of calcium represents 188 per cent of the intake, compared with a normal figure of 50 per cent, while the ratio of the urinary to the fæcal calcium is 0·84/1 compared with a normal ratio of 0·22/1.

Table I.—Calcium, Phosphorus, and Nitrogen Balance over a 3-day Period on a Low-calcium Diet

	Total Intake	Fæcal Output	Urinary Output	Balance
	mg.	mg.	mg.	mg.
Calcium	420	938	788	−1306
Phosphorus	1278	710	1036	−468
Nitrogen	7100	2940	2750	+1410

Urea-clearance tests (Van Slyke) gave figures of 99 per cent of normal in February and 60 per cent in March. Careful and frequent urinary examinations were made for the presence of Bence-Jones protein, and on two occasions, the first soon after admission to hospital and the second five months later, traces of a protein with the characteristic solubilities were suspected. There is, however, considerable difficulty in detecting traces of Bence-Jones protein in the urine in association with larger quantities of other proteins, and accordingly a 250 ml. catheter specimen was obtained, chilled immediately to 0° C. and dialysed against 0·15 molar sodium chloride solution. It was then concentrated to about 12 ml., with a protein concentration of 1 per cent, while the salt content was maintained at 0·15 molar sodium chloride. Electrophoretic examination of this preparation in a 0·2 molar phosphate buffer at *p*H 8 showed (*Fig.* 381) that there were three protein components in the urine, the fastest moving with a mobility of the order of albumin and the two slower ones behaving as γ-globulins.

Diagnosis, Progress, and Treatment.—From the radiological standpoint the diagnosis seemed to lie between osteoclastoma, enchondroma, and secondary renal carcinoma. The biochemical findings, however, were not typical of any of these lesions, but suggested rather osteitis fibrosa cystica although the normal plasma-phosphatase level is an unusual finding in hyperparathyroidism. In the circumstances it was decided to

Fig. 381.—Electrophoretic analysis of the urinary proteins.

observe the effects of radiotherapy to the sacral lesion and accordingly the patient received deep X-ray irradiation in a total dosage of 2500 r applied to the sacral area over a period of two weeks commencing April 2, 1948.

Re-examined two weeks after the course of radiotherapy the patient showed little change in his general condition although the œdema had almost disappeared on a high-protein diet given during his first stay in hospital. The radiological examinations were repeated and showed no evidence of healing of the sacral lesion while there was still no lesion discernible elsewhere in the skeleton. The biochemical findings were unchanged and the raised serum-calcium and normal plasma-phosphatase levels were confirmed on several occasions.

The generalized bone pains were not in any way relieved and it was now felt that surgical exploration of the neck was required to exclude the possibility of parathyroid adenoma or hyperplasia. This operation was performed on May 18, but after a careful search no abnormality of the parathyroid glands was detected. Further radiographs made in June showed no change in the sacral lesion, and no lesion elsewhere, so on July 1 an incision was made over the sacrum and the bone was explored. The surgeon found the cortex of the bone much thinned and giving a sensation of 'egg-shell crackling'. The centre of the lesion consisted of degenerate and hæmorrhagic tissue, fragments of which were removed for histological examination. This showed the material to consist of highly cellular and vascular tumour tissue formed of masses of rounded, oval, and more elongated cells. The majority of the cells measured between 8μ and 12μ in diameter and contained a single nucleus, but a few larger and binucleate forms were seen. The nuclei showed a well-formed nuclear membrane with the chromatin frequently aggregated into masses lying on it. A high-power view of the biopsy material is shown in *Fig.* 382. While the cells showed some of the general morphological characters of mature plasma-cells, in only a minority of them were the features quite characteristic; and the staining reactions with Pappenheim's technique were not strongly marked. In accordance with the views expressed recently by Willis (1948) the cells were regarded as neoplastic plasma-cells and a diagnosis of myeloma of plasma-cell type was recorded. Following the biopsy the patient's condition steadily deteriorated. The sacral wound failed to heal and the generalized pains were severe and persistent. Treatment with urethane and with stilbamidine, as advocated for generalized myelomatosis by Snapper (1948), brought no relief, and the œdema returned. Death occurred on July 30, eight-and-a-half years after the onset of symptoms, five-and-a-half years after demonstration of the sacral lesions in radiographs, and six months after admission to hospital.

Autopsy Findings.—Post-mortem examination was carried out six-and-a-half hours after death. External examination showed the unhealed sacral wound, which had enlarged as a result of sloughing of the skin edges.

The cardiovascular, alimentary, and endocrine systems were free from notable pathological change. The lungs showed extensive œdema of their parenchyma. The kidneys were of normal size, the left weighing 160 g. and the right 140 g.; a few capsular adhesions were present, but the cut surfaces showed normal markings. The spleen, bladder, prostate, genital organs, brain, and meninges showed no pathological features.

Examination of the sacrum revealed a large hæmorrhagic tumour of irregular outline occupying its central part. The tumour measured approximately 6 cm. in both vertical and horizontal diameters, and 4 cm. in anteroposterior thickness. The substance of the tumour was soft, pink to deep red in colour, and free from bone.

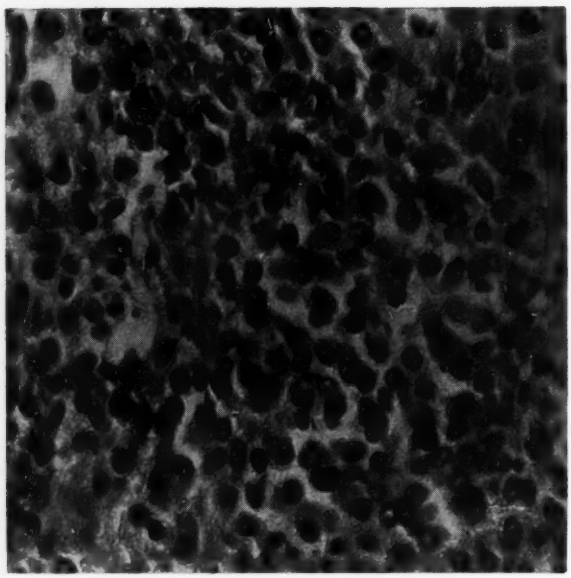

Fig. 382.—Section of the biopsy specimen. (H. and E.)

It was covered by a very thin shell of surviving bone on the anterior and posterior aspects. Careful examination of the remainder of the spine and of the ribs, skull, and long bones showed no evidence of tumour growth.

Microscopical examination of the sacral tumour showed identical features to those found at biopsy. Sections and marrow-smears were examined from many parts of the skeleton but abnormal cells were not found in any of them. Sections of the kidney showed a moderate degree of hyaline thickening of the basement membranes of many of the glomerular capillaries, producing the so-called 'platinum loop' appearance, with fixed patency. Hyaline, eosin-staining casts were seen in many of the loops of Henle, but the hyperplasia of tubular epithelium noted by Lohlein (1921) in generalized myelomatosis and by Lumb (1948) in a case of solitary tumour was not observed. Liver, spleen, and kidney were examined for amyloid deposits, but the results were negative. These gross and microscopical findings were regarded as establishing the diagnosis of solitary myeloma of the sacrum.

DISCUSSION

The diagnosis of this case presented considerable difficulty, particularly in the differentiation from osteitis fibrosa (hyperparathyroidism). The raised serum-calcium and hypercalcinuria led to a fruitless exploration of the neck for a parathyroid tumour, although it was realized that the normal phosphatase level was strongly against this diagnosis. From a radiological standpoint osteoclastoma was the most difficult alternative diagnosis to exclude, but the metabolic disturbances described are not encountered in that condition. Raised serum-calcium figures are occasionally encountered in secondary carcinomatosis of bone, but the long history in this case was against such a diagnosis. Diagnosis was in fact delayed until biopsy was performed.

The case described here seems to present many features intermediate between those of the large group of cases of myeloma which present as a localized lesion later disseminating, and those of the much smaller group of cases in which the tumour remains localized throughout a prolonged course without generalized disturbance. In this case the localized nature of the lesion has been established by post-mortem examination, and yet there were certain features usually regarded as indicative of generalization. Paul and Pohle (1940) found Bence-Jones protein in only 20 per cent (9 cases) of their series and then only when generalization had occurred. Similarly the disturbances of the serum-protein level and albumin-globulin ratio have been regarded as manifestations of the generalized disease, and this applies also to elevation of the serum-calcium with hypercalcinuria and a negative calcium balance. The case we have described shows clearly that these metabolic disturbances can occur with a true solitary myeloma. Their occurrence should not be regarded as definite evidence of generalization and the management of the case should be based rather upon an assessment of the radiological and biopsy findings and upon the accessibility of the tumour to localized excision.

The findings in this case suggest that the constitutional and metabolic disturbances are a reflection not of dissemination but of the absolute quantity of active tumour tissue present. Thus an unusually large solitary tumour may produce the constitutional and metabolic upset as effectively as a disseminated tumour. It remains a matter of conjecture as to why in the large majority of cases of myeloma the disease becomes generalized throughout the skeleton while in this smaller group it remains localized. Histologically the two types of case cannot be distinguished, though presumably there is a difference in grade of malignancy which microscopical examination fails to demonstrate.

SUMMARY

1. Solitary myeloma, remaining localized throughout its course is a rare disease. A patient with a solitary tumour of the sacrum is described.

2. Symptoms had been present for eight years and there was radiological evidence of the presence of the tumour five years and four months before death.

3. An unusual feature was the occurrence of disturbances of protein and calcium metabolism which have usually been associated with generalized myelomatosis.

4. Radiotherapy and treatment with stilbamidine were both without effect on the course and symptoms of the disease.

5. The differential diagnosis between myeloma, osteitis fibrosa, and osteoclastoma is discussed.

We wish to acknowledge the part played in this work by Dr. N. H. Martin, Chemical Pathologist at St. George's Hospital, who supervised the chemical investigations and carried out personally the electrophoretic analysis of the urinary proteins.

REFERENCES

CUTLER, M., BUSCHKE, F., and CANTRIL, S. T. (1936), *Surg., Gynec. Obstet.*, **62**, 918.
GESCHICKTER, C. F. (1930), *Ann. Surg.*, **92**, 425.
GOOTNICK, L. T. (1945), *Radiology*, **45**, 385.
KAUFMAN, J. (1945), *Amer. J. Surg.*, **69**, 129.
LOHLEIN, M. (1921), *Beitr. path. Anat.*, **69**, 295.
LUMB, G. (1948), *Brit. J. Surg.*, **36**, 16.
PAUL, L. W., and POHLE, E. A. (1940), *Radiology*, **35**, 561.
SNAPPER, I. (1948), *J. Amer. med. Ass.*, **137**, 513.
WILLIS, R. A. (1941), *J. Path. Bact.*, **53**, 77.
— — (1948), *Pathology of Tumours*, 789. London: Butterworth.

EVALUATION OF SURGICAL PROCEDURES FOR GASTRIC AND DUODENAL ULCERS*

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THE surgical treatment of gastric and duodenal ulcers has undergone constant investigation, modification, and improvement throughout the present century. Owing to the high percentage of recurrences following indirect or palliative operations, such as gastroenterostomy and pyloroplasty, these operations have been discarded. Until Dragstedt's work on vagotomy was published in 1943, adequate subtotal resection was accepted as the procedure of choice in the surgical treatment of peptic ulcer. This operation gave a high percentage of cures and a low mortality when performed by experienced and specially trained surgeons, but the occasional operator found the procedure difficult and his mortality higher than he cared to admit. Also, owing perhaps to inadequate removal of part of the stomach, recurrences were not as infrequent as desired. Certain other objections could be raised, such as the ' dumping syndrome ', in which patients continued to have distressing symptoms in spite of the absence of recurrent ulcer.

Great credit must be given to Dr. Dragstedt who, in 1943, focused attention on the value of supradiaphragmatic resection of the vagi in curing duodenal ulcers. In 1920 Ivy showed that isolated stomachs did not secrete to excess, and did not develop ulcers if freed from vagal influence. In 1930 Dragstedt demonstrated that such isolated stomachs did sometimes produce ulcers if the vagal nerves were left intact. As early as 1889 Pavlov reported that vagotomy in dogs arrested gastric secretion produced by sham feeding. Pavlov's pupil, Babkin, continued these studies at McGill University and so influenced Dr. C. A. McIntosh that he carried out two subdiaphragmatic vagotomies for duodenal ulcer in 1937. One of these recurred and continued to have free acid. I later resected this case. There is little doubt that this early attempt did not completely remove the vagi. It is impossible to review, at this time, even the important investigative work done on the influence of the vagus nerve on the gastric secretion, but for those interested I would recommend careful reading of the paper by Alvarez, " Sixty Years of Vagotomy, A Review of Some Two Hundred Articles " (*Gastroenterology*, 1948, **10**, 413). It is interesting to note that vagotomy may produce chronic gastric ulcers in dogs and rabbits. As Alvarez points out, flurries of interest have been shown in bilateral vagotomy in 1907–14, 1920–4, 1930–4, and in 1943 to the present time. The reason that surgeons have given up the operation in the first three periods has not been published.

Bilateral section of the vagus nerves undoubtedly removes psychic or neurogenic stimulation of gastric mucosa and lessens the amount of nocturnal gastric secretion. It prevents stimulation of acid secretion by hypoglycæmia and lessens night secretion. It also results in gastric atony and increases the irritability of the bowel.

The purpose of this paper is to attempt to evaluate the relative merits of these two widely utilized surgical procedures—first, subtotal gastric resection, and, secondly, bilateral vagotomy.

Those gastric surgeons who have had a wide experience have been fairly well satisfied with subtotal resections. The mortality-rate has been low, the post-operative morbidity minimal, and the hospital stay averages less than two weeks. In 1947 I carried out 100 gastrectomies for ulcer, with no mortality and an average stay in hospital of twelve and a half days. Up to Nov. 1 of this year a further 70 cases have been resected, with 1 death. This was a fortunate period as our overall mortality continues to average 2½ per cent in ulcer cases, but these cases include resection for perforated ulcer, resection for massive hæmorrhage, and one-stage operations for large calloused ulcers involving the duodenum and penetrating the pancreas. We have never refused operation no matter how serious the risk if the symptoms were of sufficient severity, or in gastric ulcers if the possibility of carcinoma could not be excluded. Such immediate post-operative complications as œdema of the stoma, which is usually due to low-grade inflammatory swelling about the suture line, have been rare. Phlebosis, embolus, and pulmonary complications have been no more common than in other major abdominal operations. Respiratory complications involved 10 per cent of the resected cases, but in only 0·3 per cent were the cause of death. Three per cent had some post-operative infection, 2 per cent developed fistulæ, slightly over 1 per cent developed œdema of the

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stoma, and the same number phlebitis. The complications were not frequent, and on the whole not often serious. With a personal experience of nearly 1000 resections, I can say that these patients are the happiest of any group I know who undergo major surgery. True, a certain percentage have symptoms characteristic of the ' dumping syndrome ' and probably as high as 3 per cent have recurrent stomach ulcer, though we have only 1 per cent of proven recurrences in this series. Inability to gain weight is also a somewhat frequent complaint.

Since our first few resections, except for minor changes, our technical procedures have been standardized. This large group forms valuable material for study. We have found, in spite of this identical technique, differing results in the later post-operative state. Quite a few have stated they cannot eat candy since this operation. In an occasional case small stomach symptoms persist for years. In another group the so-called ' dumping syndrome ' persists. This group has been relatively small in our series, perhaps because of the small stoma, or perhaps because of our psychological approach to our patients. We never let them get frightened about the operation. We treat them as normal individuals at the earliest possible post-operative day, even giving them a large steak on the ninth day. The cases of persistent dumping syndrome following resection are always found in thin, nervous, apprehensive people, usually females, who give a long history suggestive of nervous dyspepsia. By radiography or by re-operation we find no recurrence. It is true these people show nutritional disturbances, with a glucose curve which quickly rises to 200 but falls fairly rapidly to 50 or even as low as 30. I am convinced that this syndrome is a psychogenic condition occurring in neurotics, who, perhaps because of excess autonomic stimulation, developed their ulcer, not as a primary lesion but secondary to some form of neurosis, with vagal stimulation and increased gastric secretion. It is well recognized that the dumping syndrome will follow gastro-enterostomy at least as frequently as after resection.

Reviewing the situation from this large experience, it is possible to say without, I think, fear of contradiction that subtotal resection has been a most valuable addition to the treatment of ulcer. When well done it is safe, easy on the patient, and cures over 90 per cent. It is not perfect, as some post-operative symptoms and some recurrences will occur, and if a better method can be devised it should be used.

The only other surgical method which is at present considered as a probable competitor of gastric resection is vagotomy. As has been mentioned, the value of this procedure has come to the attention of surgeons on three separate occasions, only to fall into discard until 1943, when Dragstedt enthusiastically urged it for routine use in the treatment of ulcer. His thorough investigation and, for a surgeon interested in the scientific approach to surgery, his unabridged enthusiasm demanded consideration. I was not unaware of the criticism subtotal resection received when it was first proposed as the routine procedure to be followed for peptic ulcer, even though it was the first operation to be tried. In those early days, however, the mortality was too high and other safer methods such as gastro-enterostomy became the method of choice. My first reaction to Dragsted's proposal was to wait and see. I had devoted a good many years to the study and practice of resection and had, I think, had some influence on the gradual acceptance of resection for routine use for peptic ulcer. Surgeons such as Roscoe Graham, Colp, and Walters agreed with me. Subtotal gastric resection was too valuable a method to drop overnight in order to accept a new operation which had not been tried, and which in the past had been found wanting, even though it must be agreed that the earlier attempts had probably not removed all vagal fibres to the stomach. However, encouragement was given to other men in the department to try out vagotomy.

The earlier vagotomies were performed by a transthoracic approach. In spite of the enthusiasm of thoracic surgeons, and some of our younger surgeons, I am still of the opinion that a wide thoracotomy carries a higher morbidity than a simple laparotomy, and this assumption has been proven by our experience. I have permission from Dr. Campbell Gardner to report 16 cases of vagotomy which he carried out at the Queen Mary Veterans Hospital in Montreal. In addition, 22 were carried out by various members of the Royal Victoria Hospital staff—all for duodenal and stomal ulcers. I have always been opposed to the use of vagotomy for gastric ulcer on account of the danger of malignancy being present. The post-operative complications in this group were more pronounced and frequent. One death occurred from myocardial disease. In 2 cases a hæmothorax developed. Pain in the thoracic wound occurred in greater or lesser degree in almost all the cases and in some it was severe and persistent. Even the usual gastro-intestinal disturbances of gastric stasis and dilatation, feeling of fullness, and nausea were more severe. Mild and transient diarrhœa was also noted and persisted for longer periods. In all, 38 vagotomies were carried out, 21 transthoracically and 17 supradiaphragmatically. In this series of transthoracic vagotomies, 16 were reported as having good results, 4 as having poor results, and in 1 case the ulcer recurred. In the 17 cases of subdiaphragmatic vagotomy, 6 for duodenal ulcers and 11 for stomal ulcers, 9 were reported as having good results, 5 as poor results, and 3 as having recurrences. These results cover a small series carried out by several surgeons, perhaps not all complete bilateral vagotomies, and therefore it would not be sound to place too much emphasis on these figures, which are so much poorer than our figures for subtotal resection. Moore, for example, reported in 1947 40 cases with no recurrences but with undesirable after-effects (diarrhœa, fullness) in 10 per cent. He further states that at the end of a year fasting acidity and motility gradually returned to normal, though the response to the insulin test remained abolished in most cases. The clinical well-being of the patient outlasted the duration of the early secretory and motility changes. Grimson and others in 1947 reported that in 57 cases with transthoracic vagotomy alone or with gastro-enterostomy, healing of the ulcer occurred with one exception, and

persisted for from 4 to 31 months. With 8 exceptions (14 per cent) they were satisfied with the operation. They agreed further that transthoracic vagotomy had the disadvantage of not allowing for visualization of the abdominal viscera, and further where gastro-enterostomy was required because of post-operative gastric distension and retention, a second exposure was necessary. In 6 cases (10 per cent) secondary gastro-enterostomy was required. Further, 20 (35 per cent) had temporary but severe diarrhœa, and 4 patients (7 per cent) had persistent severe diarrhœa. In 6 patients (10 per cent) serious episodes of colicky abdominal pain occurred and in 1 case the duodenal ulcer persisted and was later treated by subtotal resection. Temporary difficulty in swallowing occurred in 20 patients (35 per cent). Ruffin and Smith report 30 cases of intractable ulcer with cure. In 10 per cent later gastro-enterostomy was carried out because of gastric retention. Diarrhœa (mild and transient) occurred in 40 per cent of cases. In a later report on the first 50 patients, Ruffin and others had 1 questionable recurrence and 1 death. Five others (10 per cent) were considered to have fair to poor results because of gastric retention, vomiting, diarrhœa, weakness, nervousness, or other symptoms. Eighty-six per cent were considered cured after from two to twenty-seven months—not a very long post-operative follow-up for final decisions.

The relative merits of subtotal gastric resection and vagotomy have been discussed carefully by the surgical staff of the two hospitals to which I am attached, viz., the Royal Victoria Hospital and the Queen Mary Veterans Hospital in Montreal, and we have finally come to the conclusion that vagotomy is not as useful or satisfactory an operation as resection. True, our experience has been based on a very large number of resections over many years while our experience of vagotomy is limited, but not so limited as to prevent us from having a lot of trouble with it. At present we are using vagotomy on all cases admitted with stomal ulcers following subtotal resection, and at times for ulcers recurring after a good functioning gastro-enterostomy. Where the hydrochloric acid is very high, say 90–100 units of free hydrochloric acid, a vagotomy is combined with subtotal resection, but resection has remained our standard procedure for gastric and duodenal ulcer. We are agreed that the subdiaphragmatic approach is the better. First, because we like to confirm the diagnosis by an exploratory laparotomy; and, secondly, because we are satisfied that we can do an adequate bilateral vagotomy with this approach. It is true that Dragstedt fears that a thorough vagotomy cannot always be done in this way, but we have a certain pride in our technical procedures and think that if the stomach is put on the stretch after freeing the œsophagus, all the fibres can be felt, even smaller branches. I have a certain sympathy for Dr. Dragstedt in this contention for I know, in the early days of gastric resection, how inadequate were many of the resections, how high the mortality, and how frequent the recurrences.

Our chief problems with vagotomy have been gastric atony and diarrhœa. Just recently I saw a patient six weeks after his resection and found the upper abdomen distended; on percussion an unbelievably large, distended stomach stump was found. He complained of discomfort and fullness. A check-up of the operative note revealed that he had had a bilateral vagotomy at the same time as his resection, as his free acid had risen to 100 after an alcohol test-meal. He fortunately did not have diarrhœa to add to his woes.

We realize that once a stomach is distended it seems more difficult to bring it back to normal function, so following vagotomy we keep up Wangensteen drainage for from five to seven days, which certainly is a great help but will not prevent all later trouble from distension.

Last year I decided to do some vagotomies when the free acid was high. After having done about half a dozen, my assistant resident, upon whose shoulders falls the post-operative care, begged me to do no more. The resected cases did so well, the vagotomy cases so badly. I have talked with many men who have a large series of vagotomies and who are pleased with their results—very insignificant mortality, if any; no serious trouble with distension if they keep the Wangensteen drainage up for a week and advance the post-operative feeding slowly; even diarrhœa has not been a serious problem. I can only say that this has not been our experience.

I stated before that during the last twenty months I have carried out 170 subtotal resections for ulcer with 1 death and with an average post-operative stay in hospital of twelve to thirteen days. The majority of these patients are out of bed on the first or second day, are wondering why they cannot go home at the end of six days, and are eating a steak on the ninth day. We try to get them back to work at the end of one month. With vagotomies our trouble usually starts when they should be going home. The fullness and distension of the stomach, confirmed by radiography, prevents their eating properly, and this is apt to continue for weeks. Then, if diarrhœa supervenes, we have another difficult problem to handle. In spite of our recurrences following vagotomy, which may be, and probably are, due to an incomplete operation, it is only fair to emphasize that vagotomy lowers the acid secretion, which we believe to be the major factor in ulcer chronicity, and that patients with vagotomy, when they finally get over their post-operative complications, are free from symptoms, can eat well, and tend to gain weight. There is still a certain percentage who have poor results. They continue to complain of weakness, nervousness, and indigestion, even though their pain is relieved. The literature contains sufficient evidence that the mortality is low, but recurrences have been reported with about equal frequency as after well-performed adequate resection. Undoubtedly the immediate results of subtotal resection are much superior. As the occurrence of proven stomal ulcer in our series is only 1 per cent, I have not personally felt it necessary or wise to combine vagotomy with resection except in cases showing a very high free acid. It appears to me to be wiser to perform a vagotomy only on those few cases which later develop a stomal ulcer. On the other hand, with vagotomy, gastric retention is such a frequent post-operative complication that I think a resection,

gastro-enterostomy, or pyloroplasty should be added to vagotomy.

Vagotomy should not be carried out for gastric ulcer because of the large percentage of mistaken diagnoses. Last year I had 4 cases presumed to be benign peptic ulcer, as judged by clinical and radiographic criteria and examination of the gross specimen, which proved to be carcinoma on careful histological scrutiny. This is an unusual high frequency of such cases, but demonstrates the danger of doing only a vagotomy in cases of gastric ulcer. In the case of acute perforation it hardly seems wise to open the mediastinum in the presence of contamination of the abdominal cavity. In cases perforated under twelve hours I usually carry out a subtotal resection, in later cases I simply close the perforation, or cover it with omentum.

Vagotomy might be satisfactory in cases of massive hæmorrhage if the bleeding vessels are ligated or the ulcer, if gastric, is excised. But as such cases will so frequently need a gastro-enterostomy, either at the time of operation or later, I feel a subtotal resection to be the method of choice.

I would like to report one interesting death following vagotomy. A male patient, 50 years of age, had dumping syndrome, with pain, for sixteen years following gastro-enterostomy. Though no ulcer was demonstrated by X rays, a laparotomy was carried out on account of the severity of the symptoms. No duodenal or stomal ulcer was found, so a vagotomy was performed in the hope of arresting the dumping syndrome. The operation was made easy by the thinness of the patient. He did well for two days, but then developed an increasing paralytic ileus despite Wangensteen drainage. Two weeks later the abdomen was again explored and the small and large bowel were found to be distended down to the splenic flexure but no mechanical obstruction, or peritonitis, was found. A cæcostomy was carried out to relieve distension but this was ineffectual. The patient died twenty days post-operatively. Autopsy revealed no mechanical obstruction, death being ascribed to ileus. Though this may be an unexplainable case of paralytic ileus, it would appear possible that the section of the vagi had so upset the autonomic nervous balance of the gastro-intestinal tract, that it played a part in the initiation and continuance of the ileus. The anatomic distribution of the distended bowel corresponded to the vagus distribution. It is interesting that in 1000 cases of subtotal resection performed personally no case has died from idiopathic paralytic ileus.

In conclusion it may be stated that though subtotal gastric resection may not be the perfect operation for gastroduodenal ulcer, in our experience it has proved to be the most satisfactory. The mortality is low (2·5 per cent), the post-operative course comparatively free from serious complications, the patients are able to go home on the tenth post-operative day, usually free from symptoms, eating well, and feeling fine. The proven recurrences are less than 1 per cent in our series. For these recurrences we have found vagotomy very satisfactory in a very short-term follow-up.

Our experience with vagotomy is unsatisfactory because of the post-operative complications of distension and diarrhœa, and because deaths and recurrences have occurred in our rather small series. Subdiaphragmatic vagotomy is simple and perhaps should be used by the occasional operator whose training and experience makes a subtotal gastric resection too hazardous for the patient. Vagotomy, however, appears to be the operation of choice for stomal ulcer following gastric resection, and, combined with resection, for those ulcer cases with a free acid reaching as high as 90–100. I cannot, at this time, recommend it as a routine procedure for duodenal ulcers in place of gastric resection and believe it should never be used for gastric ulcers.

LYMPHANGIOMA OF MESENTERY

By IAN MACNAB and T. MENZIES

Tumours of the mesentery are not common and, when seen, present such a bizarre pathological picture that one is tempted to regard them as unique surgical lesions. However, a cursory glance through the literature reveals that several hundred cases have previously been reported.

Cysts of the mesentery were first described in 1507 by Benevieni, a Florentine anatomist. No further mention of them seems to have appeared in medical literature till 1803, when Portal made an attempt to classify them. Rokitanski in 1842 was the first to describe a chylous cyst of the mesentery, but it was not until 1880 that the first cyst was successfully operated on by Tillaux.

Warfield (1932), after an exhaustive search through the literature, estimated that 500 cases of mesenteric cysts of all types had been described. Since the publication of his paper, about 200 cases have been recorded, making the total number to date approximately 700 cases.

Lymphatic tumours of the abdomen, however, are very uncommon. Watson and McCarthy (1940) reported on 1056 tumours of blood-vessels and lymphatics seen between 1931 and 1939; they found only 41 lymphangiomata, none of which were intra-abdominal, and Rayford (1932) stated that he found only 1 lymphangioma of the abdominal viscera in 11,500 post-mortems. Intra-abdominal lymphangiomata occur most commonly in the great omentum and it is extremely rare for them to develop in the mesentery. In view of this, the following case is of interest.

CASE REPORT

Recruit E. E., aged 18, was admitted to the Cambridge Military Hospital, Aldershot, on March 21, 1948. On the night before admission he had developed colicky suprapubic pain. The pain had gradually increased in severity and by that morning had settled in the left iliac

fossa. He vomited once only. His bowels were regular and he had no urinary symptoms.

On Examination.—His temperature was 101° F. and his pulse-rate was 120. He was flushed, but his tongue was clean and moist. There was marked guarding and tenderness in the left iliac fossa, but nothing else abnormal was detected. There was no dextrocardia.

It was decided that he should be treated conservatively. He was given an enema which produced a good result and gave considerable relief of symptoms. The tenderness in the left iliac fossa, however, persisted. He was started, empirically, on a course of sulphamezathine. Over the next 48 hours his temperature rose to 102° and on March 23 the pain became more severe and began to spread to the right iliac fossa. It was decided that an exploratory laparotomy should be performed.

At Operation (I. M.).—The abdomen was opened by a paramedian subumbilical incision. There was a small amount of serous effusion in the peritoneal cavity. About 2 ft. of ileum, 18 in. away from the ileocæcal valve, was enveloped by a mass of cyst-like structures. Superficially the condition was very similar in appearance to diverticulitis of the colon. The cysts had opaque, well-defined walls, and it appeared that an acute inflammatory process had started in some of them. The mesentery was grossly thickened, œdematous, and hyperæmic. There were many enlarged mesenteric lymph-nodes showing petechial hæmorrhages on their surfaces. A similar cystic change, but to a lesser degree, was present in the meso-appendix. No other cysts were found in the abdomen.

In view of the hyperæmic and œdematous state of the mesentery, it was decided that primary resection of the loop of bowel was too dangerous a procedure to undertake. A lateral anastomosis of the ileum above and below the affected segment was performed and the bowel marsupialized (*Fig.* 383). Appendicectomy was performed.

Fig. 383.—The marsupialized portion of bowel.

Post-operative Course.—The patient was transfused with two pints of blood and one pint of glucose-saline following the operation. A week later the marsupialized loop of bowel was resected. His general condition continued to improve, but after five days he developed a fæcal fistula. This did not close spontaneously and required operative closure a month later. He is now completely relieved of all his symptoms.

Pathological Report on Specimens.—

a. Macroscopically.—The specimen consisted of 2 ft. of ileum. The antimesenteric border was free and showed no abnormality. In the mesentery, close to the ileum and abutting against the intestinal wall, there were a number of cyst-like structures enveloping the intestinal circumference. There were about twenty-two large cysts present, varying in size from 1 cm. to 10 cm. in diameter. They were adherent to each other by a layer

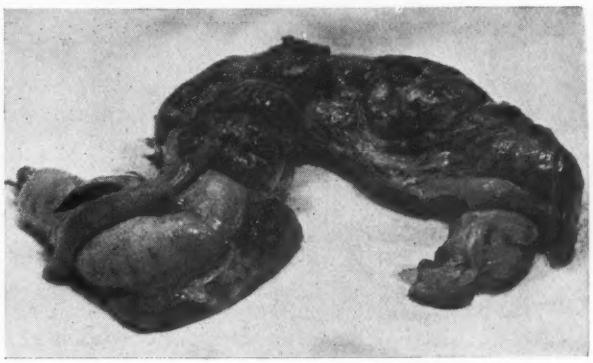


Fig. 384.—The resected loop of bowel. The cysts have been matted together by a fibrin deposit and are not clearly defined. The abrupt change from the normal ileum to the affected portion at either end of the specimen is clearly shown.

of fibrinous exudate but could be separated, and the lines of separation ran at right angles to the long axis of the bowel. The cysts were sessile; some were soft and some tense. Many of the cysts intercommunicated and there had been hæmorrhages into some of them. At either end of the specimen there

Fig. 385.—Radiograph of the affected loop filled with barium. This shows that the cysts do not communicate with the bowel lumen.

were two or three inches of normal ileum. The change from normal to abnormal was quite abrupt (*Fig.* 384).

The mucous membrane of the ileum was apparently normal. At the junction of the lower and middle

thirds, to an extent of about 4 in., there was a darkish discoloration due to hæmorrhage showing through the mucous membrane.

There was no evidence of any communication between the cysts and the mucous lining of the intestine. To demonstrate this fact, the resected loop was filled with barium and a radiograph was taken (*Fig.* 385). The cysts did not fill with barium. Some barium was then injected into the larger cysts to show their relation to the lumen of the bowel (*Fig.* 386). The barium was

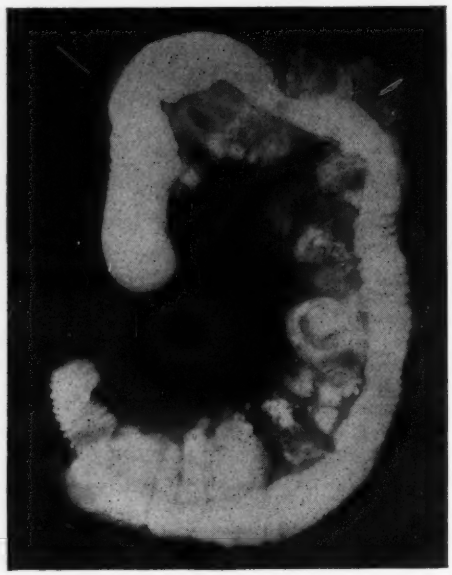


Fig. 386.—Radiograph of the affected loop with barium injected into the larger cysts to show the relation of the cysts to the bowel wall.

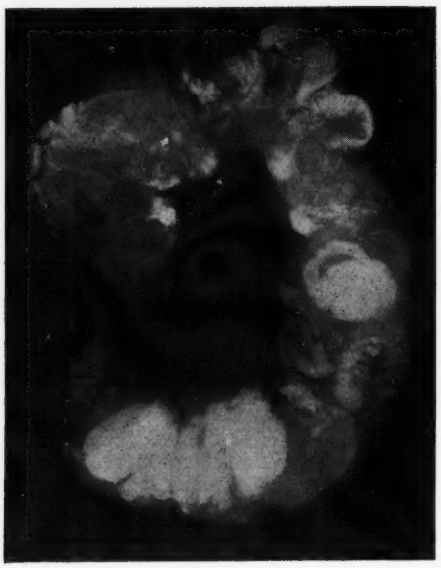


Fig. 387.—The bowel has been emptied, but barium remains in the cysts.

then washed out of the bowel and a further radiograph showed that the cysts still retained the barium which had been injected into them (*Fig.* 387).

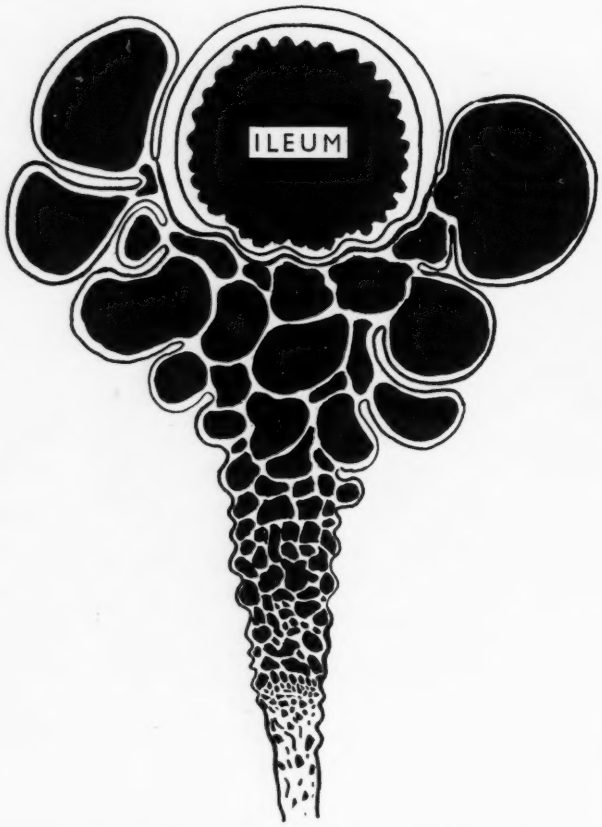


Fig. 388.—A diagram of a section through the bowel and mesentery, showing the relationship of the larger cysts to the bowel wall, and the honeycomb arrangement of the cysts in the root of the mesentery.

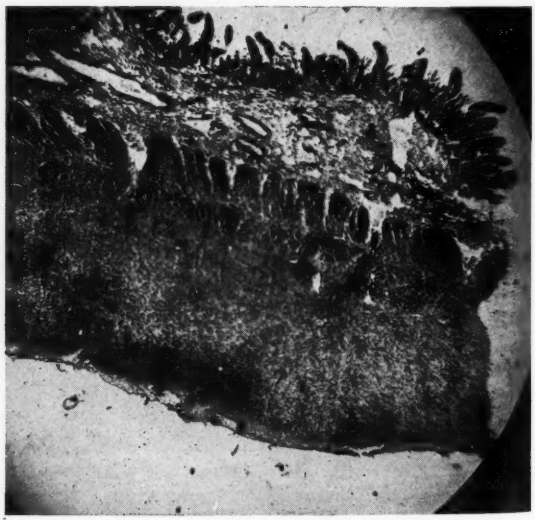


Fig. 389.—Low-power view showing the cystic spaces in the submucosa. (× 15.)

Some cysts contained brownish, blood-stained fluid, some were filled with blood-clot, and others contained clear straw-coloured fluid. The walls of the cysts were about 1 mm. thick and the lining was smooth and glistening and showed marked trabeculation. Almost every cyst was multilocular, some showed a few relatively large spaces, others were honeycombed. This honeycombed appearance became more marked towards the mesentery, which, on being cut, was seen to consist of a maze of small intercommunicating cysts (*Fig.* 388).

b. Microscopically.—The striking feature was the cystic nature of the mucous membrane (*Fig.* 389).

These cystic spaces replaced most of the normal mucosa and submucosa and, in places, there was only one layer of cells separating them from the lumen of the bowel. The muscularis mucosa and the submucous tissue were destroyed and replaced in great part by these cysts, which were lined with flattened, non-epithelial cells. The cysts were separated by fine, loose areolar tissue in which lymphocytes were common. The contents of the cysts were a fine, meshwork coagulum containing lymphocytes. Mucous membrane cells were in great part deficient, and were only seen in the crypts (*Fig.* 390).

Fig. 390.—Cystic spaces are seen to replace most of the mucosa and submucosa. The cysts are lined by flattened non-epithelial cells. The cysts contain a fine meshwork coagulum, in which are seen several lymphocytes. (× 75.)

The walls of the larger cysts consisted of loose, cellular, fibrous tissue interspersed with small areas of adipose tissue, liberally endowed with vessels and containing a large number of lymphocytes. At the internal surface there was a condensation of these structures and the lining was of the same flattened, non-epithelial cellular type. Lymphatic vessels were numerous in the walls, and, in many places, showed cyst-like dilatations.

There was hæmorrhage into some of the cysts, and in some of the clots there was evidence of an acute inflammatory process having taken place.

In certain areas of the thicker portions of the cyst walls, there were small collections of smooth muscle-bundles separated by a luxurious network of small lymphatic vessels containing numerous lymphocyte-like and monocytoid cells.

The mesentery exhibited the broad structural arrangement outlined above—a loosely textured, vascular, fibro-areolar tissue, showing numerous irregular spaces, all imperfectly lined with a flattened, non-epithelial type of cell (*Fig.* 391). Here, too, there was organizing clot in some of the spaces, and, in one coagulum, there was definite evidence of pyogenic infection.

The one striking histological difference between the mesenteric honeycomb and the larger, more peripheral, cysts was the relative acellularity of the mesentery. Lymphocytic cells, single and in collections, and monocytoid and fibrous tissue cells, were present to a marked degree, but were fewer in number than in the portions nearer the ileum (*Fig.* 392).

CONCLUSION.—The overall picture—cysts lined by imperfect, flattened, non-epithelial types of cells, and containing a yellowish fluid, numerous lymphocytes, and monocytoid cells; the abundance of vessels; the cellularity of the walls, the presence in them of smooth muscle and of all gradations of lymph-spaces—points to the nature of the condition being that of a lymphangioma.

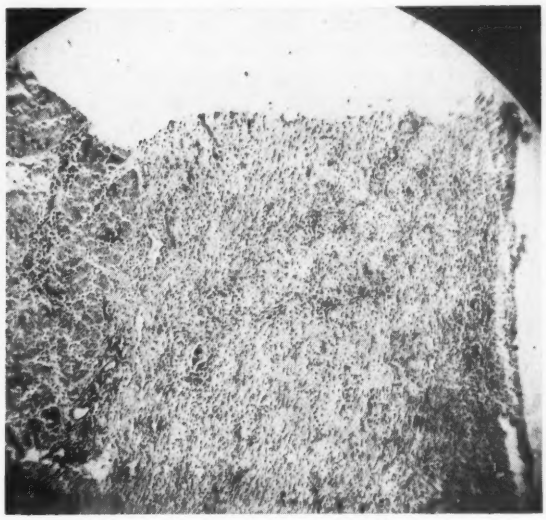


Fig. 391.—Section of the juxta-ileal part of the mesentery, showing the dense collection of lymphocytes and monocytoid and fibrous tissue cells. (× 45.)

DISCUSSION

AETIOLOGY

There is considerable doubt as to whether these tumours are all true angiomatous formations. Carter (1921) stated that they were derived from

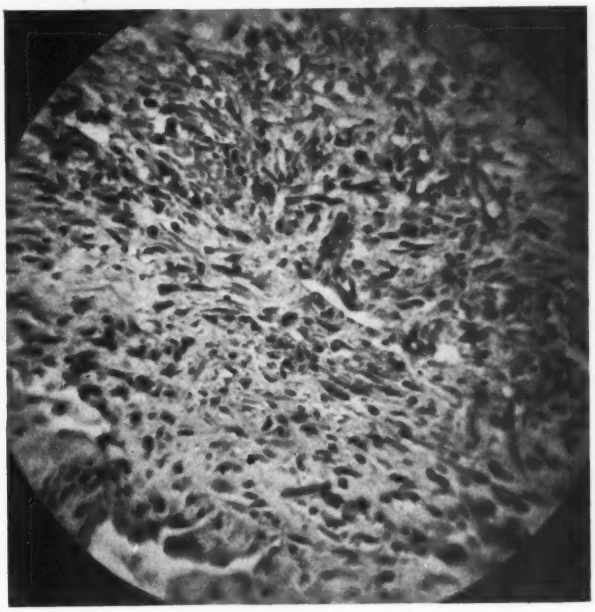


Fig. 392.—High-power view of the dense collection of cells in the mesentery. Note the dilated lymph-spaces. (× 195.)

the remnants of a Meckel's diverticulum, and it is interesting to note that, in the case reported above, the cystic change occurred about 2 ft. from the ileocæcal valve. They have also been described as being dilatations of lymphatic vessels (Moynihan, 1897), and as degenerated lymph-nodes (Rokitanski, 1842).

Some pathologists hold the view that these tumours arise as the result of stasis of the efferent channels of the abdominal lymphatics, and that the proliferative changes which are seen are due to the irritative effects of pressure. Wegner filled the peritoneal cavity of animals with air and maintained this increased abdominal pressure for some time. Air entered the subperitoneal lymphatics and the lymphatics of the great omentum. This produced an obstruction of the efferent vessels leading to the development of large cysts with proliferative changes in their walls. These experiments are of particular interest in view of the increasing use of artificial pneumoperitoneum in cases of phthisis. Ewing (1928) supported the view that these cysts were developed by congenital or acquired obstruction of the mesenteric lymphatics. Ribbert (1898), however, denied that stasis played any part in their production, and Dowd (1900) was of the opinion that the extensive anastomoses of the lymphatic plexuses of the intestines and mesentery precluded obstruction.

Sick (1902, 1903) and Borchers (1920) believed that stasis plays a secondary role, and leads only to the enlargement of the tumour.

The consensus of opinion is that these cyst-like structures are true tumours which most probably arise from misplaced embryonal nests (Crane, 1930; Grausman and Jaffe, 1928), or from post-natally developed connective tissue, having the power to produce lymph-channels. Enlargement of the tumours is probably due to lymphatic stasis.

PATHOLOGY

Macroscopically lymphangiomata may present as single unilocular cysts, single multilocular cysts, or in a polycystic form. They may reach an enormous size; the largest reported was 75 lb. in weight (Ormsby, 1883).

The microscopical features of the case previously reported are similar to those found in this patient.

The cysts are lined by a flattened non-epithelial type of cell, and contain a meshwork coagulum in which there are many lymphocytes. Most authors agree that the walls of the larger cysts contain smooth muscle and a dense network of lymphatic vessels.

The cysts may be separated by loose areolar tissue or dense cellular tissue containing many lymphocytic type of cells. In some cases the intervening tissue consists of myxomatous cells.

CLINICAL PICTURE

Mesenteric lymphangiomata do not produce a constant clinical picture. They may present as an abdominal catastrophe, with severe pain, vomiting, and abdominal rigidity, or as an ill-defined malaise, the patient complaining of "weak blood" as in the case reported by Collins and Berdez (1934).

The history of each case is determined by the complications which have occurred. Waters (1946) states that one-third of the cases produced intestinal obstruction. The tumours may undergo torsion or produce an intestinal volvulus. They may rupture into the peritoneal cavity and simulate acute peritonitis or a ruptured tubal gestation. There may be slow hæmorrhage into the cyst over a long period, producing the picture of secondary anæmia. Frequently they become infected and present as an acute appendicitis or diverticulitis, and this is what apparently happened in the case we report. Perhaps most commonly they are discovered accidentally during an exploratory laparotomy.

SUMMARY

1. Another case of lymphangioma of the mesentery is added to the few previously recorded.

2. The aetiology of the condition is discussed.

3. The clinical picture, as shown by the previous cases reported, depends on the complications that have occurred.

We would like to thank Brig. Fettes, Director of Surgery for the Army, for permission to publish this case; Lt.-Col. Sachs, R.A.M.C., for the photomicrographs; and Major Lynch, R.A.M.C., for the report on sections from the specimen.

REFERENCES

BENEVIENI (1507), quoted by WARFIELD.
BORCHERS, E. (1920), *Beitr. klin. Chir.*, **118**, 710.
CARTER, R. M. (1921), *Surg., Gynec. Obstet.*, **32**, 892.
COLLINS, A. N., and BERDEZ, G. (1934), *Arch. Surg.*, **28**, 325.
CRANE, W. (1930), *Amer. J. Surg.*, **9**, 441.
DOWD, C. N. (1900), *Ann. Surg.*, **32**, 515.
EWING, J. (1928), *Neoplastic Diseases*, 3rd ed. Philadelphia: W. B. Saunders Co.
GRAUSMAN, P. M., and JAFFE, H. L. (1928), *Ann. Surg.*, **87**, 66.
MILLS, W. G. Q. (1941), *Brit. J. Surg.*, **29**, 275.
MOYNIHAN, B. G. A. (1897), *Ann. Surg.*, **29**, 1.
MURBACH, C. F., LEWISON, E. F., and DEIBERT, G. A. (1945), *Amer. J. Surg.*, **68**, 391.
ORMSBY, G. (1883), *Med. Pr.*, **25**, 258.
PORTAL, R. (1803), quoted by WARFIELD.
RAYFORD, T. S. (1932), *Arch. Surg.*, **25**, 122.
RIBBERT, E. (1898), *Virchows Arch.*, **151**, 381.
ROKITANSKY (1842), quoted by MOYNIHAN.
SICK, C. (1902), *Virchows Arch.*, **170**, 9.
— — (1903), *Ibid.*, **172**, 445.
TILLAUX (1880), quoted by WARFIELD.
WARFIELD, J. O. (1932), *Ann. Surg.*, **96**, 329.
WATERS, E. G. (1946), *Amer. J. Obstet. Gynec.*, **52**, 478.
WATSON, W. L., and MCCARTHY, W. O. (1940), *Surg., Gynec. Obstet.*, **71**, 569.
WEGNER, quoted by GRAUSMAN and JAFFE.

CONGENITAL ABSENCE OF THE SACRUM AND COCCYX

REPORT OF A CASE AND REVIEW OF THE LITERATURE

By BERNARD FREEDMAN, London

Congenital absence of part or whole of the sacrum is rare. Sporadic cases of this interesting anomaly have been reported in the past, but there has so far been no comprehensive account of the condition. Agenesis of the caudal end of the spine may vary in degree from absence of all vertebræ below the lower thoracic region at the one extreme, to a mere absence of the terminal portion of the coccyx at the other. Gross agenesis of the caudal portion of the spine, from mid-thoracic level or higher, is probably incompatible with life at an early stage of fœtal development and would therefore tend to remain unreported. Absence of the terminal part of the coccyx would also tend to remain unreported owing to the lack of any symptoms leading to its discovery. Recorded cases, therefore, probably represent only those of intermediate severity. Forty-four cases of congenital absence of part or whole of the sacrum and coccyx have been hitherto placed on record since the condition was first described by Hohl in 1852, and this paper contributes one other. The essential features of these cases are outlined in *Table I*, in which they are grouped according to the level of the lowest vertebral segment present. It will be seen that they form a continuous gradation of degrees of agenesis, beginning with a case in which the vertebral column was absent below the tenth thoracic segment, and ending with one in which merely three of the four coccygeal segments were lacking. In determining the level of the lowest segment present there may in some instances have been an error of one, perhaps two, segments. This is due to the fact that the lowest segments may be cartilaginous, or to fusion of adjacent vertebræ. Where the examination depends on radiology, cartilaginous segments may be missed yet subsequent autopsy would show them to be present.

AETIOLOGY

Theories as to causation suggested by previous writers have been based on a belief in the suppression of metameric segments, but go no further than that.

In all recorded cases there was total absence of the vertebral column below a certain level. This suggests that the cause, whatever it may be, operates from below upwards. The fact that the lowest vertebræ present are sometimes malformed or rudimentary further suggests that at this slightly higher level its inhibitory influence operates less strongly; and this, too, lends support to the hypothesis of a noxious factor operating from below upwards. Brack's case (1929) in a human stillborn child is reminiscent of the pigs without hind limbs, mentioned below, and may represent a spread of the inhibitory process. Here the lower limbs appeared on external examination to be completely absent. Radiologically a left femur was seen, and there were indications of a vestigial left tibia and right femur.

It is certain that the defect occurs in the early stages of fœtal development, and the present writer's suggestion is that this anomaly is due to interference with a primary organizer. Suppression of evocators has been achieved experimentally. Thus rumpless chicks, lacking synsacrocaudal vertebræ and the five free caudal vertebræ, have been produced by subjecting the developing eggs to high temperatures (Danforth, 1932), lithium salts have caused suppression of the notochord (Lehmann, 1935), and deficiency of vitamin A in the diet of sows has resulted in the birth of pigs without hind limbs (Zilva, Drummond, and Coward, 1921). Conceivably illness, pyrexia, or the presence of toxic substances in the maternal circulation might similarly interfere with an organizer or suppress an evocator in a human embryo. The possibility of this disorder being due in some cases to a genetic mutation is dealt with later in a discussion of the hereditary aspects.

CLINICAL FEATURES

Vertebral and Pelvic Abnormalities.—Any degree of agenesis of the vertebral column may be present, from absence of the lower portion of the coccyx alone, to complete lack of spine below the tenth thoracic vertebra. Frequently the lowest two or three vertebræ present are small and malformed or even rudimentary. Functionally these cases may be divided into two broad classes. In one, the first sacral segment is present, or, failing that, the fifth lumbar vertebra is sacralized. The bony ring of the pelvis is then complete and the weight of the trunk is transmitted normally, or almost so, through the innominate bones to the femora. Stance and gait are then almost or quite normal. In the other functional category, the sacrum is totally missing and no sacralized lumbar vertebra is present to do its work. The iliac bones then meet in the midline posteriorly, the buttocks are flattened, the natal cleft is short or absent, and the transverse diameter and outlet of the pelvis are much diminished. Hamsa (1935), Zeligs (1940), and Foix and Hillemand (1924) show illustrations of the characteristic external appearances. In the severer grades standing and walking are impossible. Since the iliac bones in such instances are in contact, they assume a vertical position and dislocation of the hips usually results. Associated abnormalities recorded are sacralization of the lowest lumbar vertebra present (Foix and Hillemand, 1924; Kienböck and Zimmer, 1935; Lichter, 1947), spina bifida (Hamsa, 1935; Zeligs, 1940; Charlier and Sauty, 1913; Hilgenreiner, 1937; Lichter, 1947; Hohl, 1852), hemivertebræ (Zeligs, 1940; de Araújo, 1936), wedge-shaped lumbar vertebræ in numerous instances often causing scoliosis, wedge-shaped lower cervical and upper thoracic

Table I.—CASES CLASSIFIED ACCORDING TO LEVEL OF LOWEST VERTEBRA PRESENT, WITH INFORMATION CONCERNING THE COMMONER ASSOCIATED CLINICAL FINDINGS

LOWEST VERTEBRA PRESENT	AUTHOR	AGE ON EXAMINATION	SEX	ALIVE OR DEAD ON EXAMINATION	URINARY INCONTINENCE	FÆCAL INCONTINENCE	CLUBFOOT	HIP DISLOCATION	MUSCULAR WASTING	DATA CONCERNING VISCERA, ETC.
Thoracic 10	Friedel (1910)	Birth	F.	D			+			Vagina rudimentary
Thoracic 11	Sinclair and Duren (1941)	9 yr.	M.	A	+	+	*	*	+	* Severe deformities of lower limbs
Lumbar 1	Feller and Sternberg (1931)	SB	F.	D			+			Rectal atresia
Lumbar 2	White (1911)	SB	M.	D			+			One median kidney. L. ureter vestigial. Rectum absent. R. vas absent. R. testis ectopic
Lumbar 3	Hamsa, I (1935)	14 mth. and 9 yr.	F.	A	+	+	+	+	+	
	Desfosses and Mouchet (1924)	7 yr.	F.	A	+		+	−		Rectum opens into vagina
	Drehmann (1927)	1 yr.	M.	A			−	+		
	Pouzet, I (1938)	15 dy. and 3 yr.	M.	A	−	−	+	+	+	Son of Pouzet's case II
Lumbar 4	Hamsa, II (1935)	9 yr.	M.	A	+	±	+	+	+	Testis rudimentary
	Zeligs, I (1940)	7 mth.	F.	A			+	−	+	
	Brailsford (1929)	5 yr.	M.	A	+	+	+	−	+	
	Elmslie (1921)	14 mth.	M.	A	(+)	+	+	+		Patent foramen ovale
	Higgs (1924)			D						
	Espasa (1928)	7 yr.	M.	A	+	±	+	−	+	
Lumbar 5	Wertheim (1857)	8 dy.	F.	D						
	Mally (1903)	9 yr.	M.	A	+	−	+	+	+	Testis ectopic
	Charlier and Sauty (1913)	4 mth.	M.	A	(+)	(+)				
	Stewart (1924)	6 yr.	M.	A			+			
	Leri and Linossier (1925)	41 yr.	F.	A	+		+			2 SB children
	Brack (1929)	SB	M.	D						Absent external genitals. Vestigial lower limbs
	Girard (1935)	6 yr.	F.	A	+	+	+	−		Rectum and vagina enter cloaca
	Hilgenreiner (1937)	2½ yr.	F.	A	−	−	+	+	+	Anal stricture
	Berman (1945)	20 yr.	F.	A	+	±	−	−	+	1 child with C.H.D., Cæsarean
	Price (1933)	16 yr.	F.	A	+	+	−	−	+	Complete transposition of viscera
	Lichter (1947)	8 mth.	F.	A	(+)	(+)	+			Fused kidneys, hydrocephalus
	Bowman (?)	6 yr.	M.							
Sacral 1	Fitch (1910)	6 yr.	M.	A	+	±	−	−	+	
	Rendu and Verrier (1913)	1½ yr.					−	+	+	Hypospadias
	Bradburn (1922)	2 yr.	M.	A						
	Foix and Hillemand (1924)	51 yr.	M.	A	+	−	−	−	+	
	Müller (1936)	6 yr.	M.	A	+	+	+	−	+	
	Pouzet, II (1938)	30 yr.	M.	A	−	−	−	−	+	Father of Pouzet's case I
Sacral 2	Albrecht (1885)									
	Litzmann (1884)	36 yr.	F.	A	+	−	+	−	−	3 children
	Foix, Achard, and Mouzon (1924)	21 yr.	F.	A	+	−	−	−	+	Bladder diverticulum
	Zeligs, II (1940)	5 yr.	F.	A	−	−	+	−	−	Rectum, partial atresia. Urethra opens into vagina
	Guilleminet (1938)	12 yr.	M.	A	−	±	−	−	+	
	Diaz Lira (1938)	9 yr.	M.	A	+	−	+	−	+	
	Hohl (1852)	SB	F.	D						Absent L. Fallopian tube. Absent L. kidney. Double R. ureter. Bilateral ovarian cysts. Absent R. hypogastric and R. umbilical arteries
	Freedman (1950)	61 yr.	M.	A	+	−	−	−	+	L. hydronephrosis
Sacral 3 or 4	Lamoot and Graux (1936)	8 yr.	F.	A	+	+	−	−	+	
Sacral 5	Newell (1934)		F.	A						R. kidney absent. Uterus unicornis rudimentary
Coccygeal 1	Kienböck and Zimmer (1935)	51 yr.	M.		−	−	−	−	−	

Barros Lima (1935), Pérez Castro (1944), and de Araújo (1936) have each reported one case, but the necessary particulars for inclusion in the table are not available.

A = alive. D = dead. SB = stillborn.

(+) denotes that patient was too young for incontinence to be of significance.

I and II refer to different cases described by the same author.

vertebral bodies (Fitch, 1910), meningocele (Charlier and Sauty, 1913; Lichter, 1947), fusion of adjacent ribs (Hamsa, 1935; Zeligs, 1940; Hohl, 1852), and articulation of opposite ribs in the midline where the corresponding vertebra is absent (Sinclair and Duren, 1941).

Neurological Abnormalities.—These result from interference with or agenesis of the spinal nerves in the affected segments. The following are the consequences :—

a. Incontinence of urine, which was present in the vast majority of reported cases.

b. Incontinence of fæces is rarer, and tends to improve at puberty. Even in the absence of fæcal incontinence, the anal sphincter is usually atonic.

c. Atrophy of muscles supplied by spinal nerves of the affected segments. This is doubtless muscular hypoplasia or agenesis, rather than atrophy. They are small or clinically undetectable. The muscles mainly involved are those of the perineum (S.2–5), legs and buttocks (L.5–S.1), while most of the thigh muscles (quadriceps and adductors, L.2–4) are normal. This results in characteristically cone-shaped lower limbs, and contributes to the flattening of the gluteal convexity with absence of the natal cleft, already noted as being due in part to skeletal changes. It is also responsible for the incontinence.

d. Sensory changes occur, but less frequently than motor changes, and their segmental distribution appears to be smaller. The comparative infrequency of reports of sensory loss may be partly due to the infancy of the patient in many instances, or to incomplete examination.

e. Deformities of the lower limbs occur very frequently and are mostly of paralytic type. Club-foot is present in half the cases. It may be equinus, calcaneus, valgus, or varus, alone or in various combinations. Genu recurvatum is mentioned twice (Hamsa, 1935; Hilgenreiner, 1937). The frequent occurrence of dislocation of the hips due to vertical iliac bones may be mentioned again in connexion with deformities; it is recorded eight times (18 per cent).

Visceral Abnormalities.—These are listed in the table. They were reported in 16 cases (35 per cent) and the genito-urinary system or rectum was involved in all but one of these, the odd one being a case of transposition of the viscera without other evidence of structural abnormality of the viscera. The true incidence is doubtless higher, since in some cases discovery of the abnormality is dependent upon autopsy and most of the cases reported were alive at the time of examination. The limitation of these anomalies to the pelvic organs and parts derived therefrom may be causally related to the spinal defect affecting the same region, the inference being that the same noxious influence may affect the hind-gut, cloaca, and the Wolffian and Müllerian ducts. However, this point must not be unduly stressed, since, of the visceral abnormalities that are sometimes associated with, for example, congenital absence of the tibia, many are ano-genito-urinary, anal atresia, hermaphroditism, and hypospadias having been described (Dankmeijer, 1935).

Three affected women bore children (Sinclair and Duren, 1941; Feller and Sternberg, 1931; Berman, 1945). Labour was difficult in all cases, one dying at the birth of her third child (Feller and Sternberg, 1931), and another requiring Cæsarean section (Berman, 1945). There is no information concerning potency and fertility in affected adult males, with the exception of the patient who succeeded in producing a son with the same sacro-coccygeal anomaly (Pouzet, 1938). The secondary sex characters are normal, and in the absence of any information to the contrary, it may probably be taken that function too is normal.

Sex Incidence.—The sex has been recorded in 39 cases; of these 22 were male and 17 female. There is thus a slight preponderance of males, the ratio being 1·3 : 1. This bears comparison with the ratio of 1·6 : 1 for congenital absence of the tibia (Dankmeijer, 1945), and with the preponderance of males that exists for most other congenital abnormalities.

Inheritance.—There is a single instance of congenital absence of the sacrum occurring in more than one member of the same family, father and son (Pouzet, 1938), and a familial tendency may have existed in Zelig's second case (1940), in which one sibling of a total of six had a meningocele, and the mother's sister and father's brother each had a child with congenital club-feet. Here a combined tendency from both parents may have existed. With regard to the mechanism of inheritance, and the writer's suggestion that the condition may be due to interference with an organizer, Needham (1942) has made some interesting speculations on the possible relationship between evocators and genes.

Rumplessness in fowls, with its associated lack of synsacrocaudal vertebræ, the five free caudal vertebræ, the uropygial oil gland, and frequent associated visceral abnormalities, has already been referred to in connexion with its experimental production. This condition, known for three centuries (Highmore, 1651), closely resembles structurally the human disorder which is the subject of this paper; it occurs both as a hereditary anomaly (Landauer, 1933; Dunn and Landauer, 1934, 1936), and spontaneously (Dunn, 1925). It may also occur in varying degrees of severity (Landauer, 1928). With so many points of resemblance it is tempting to assume that an analogous process is at work in some human cases by an inherited or mutant gene.

TREATMENT

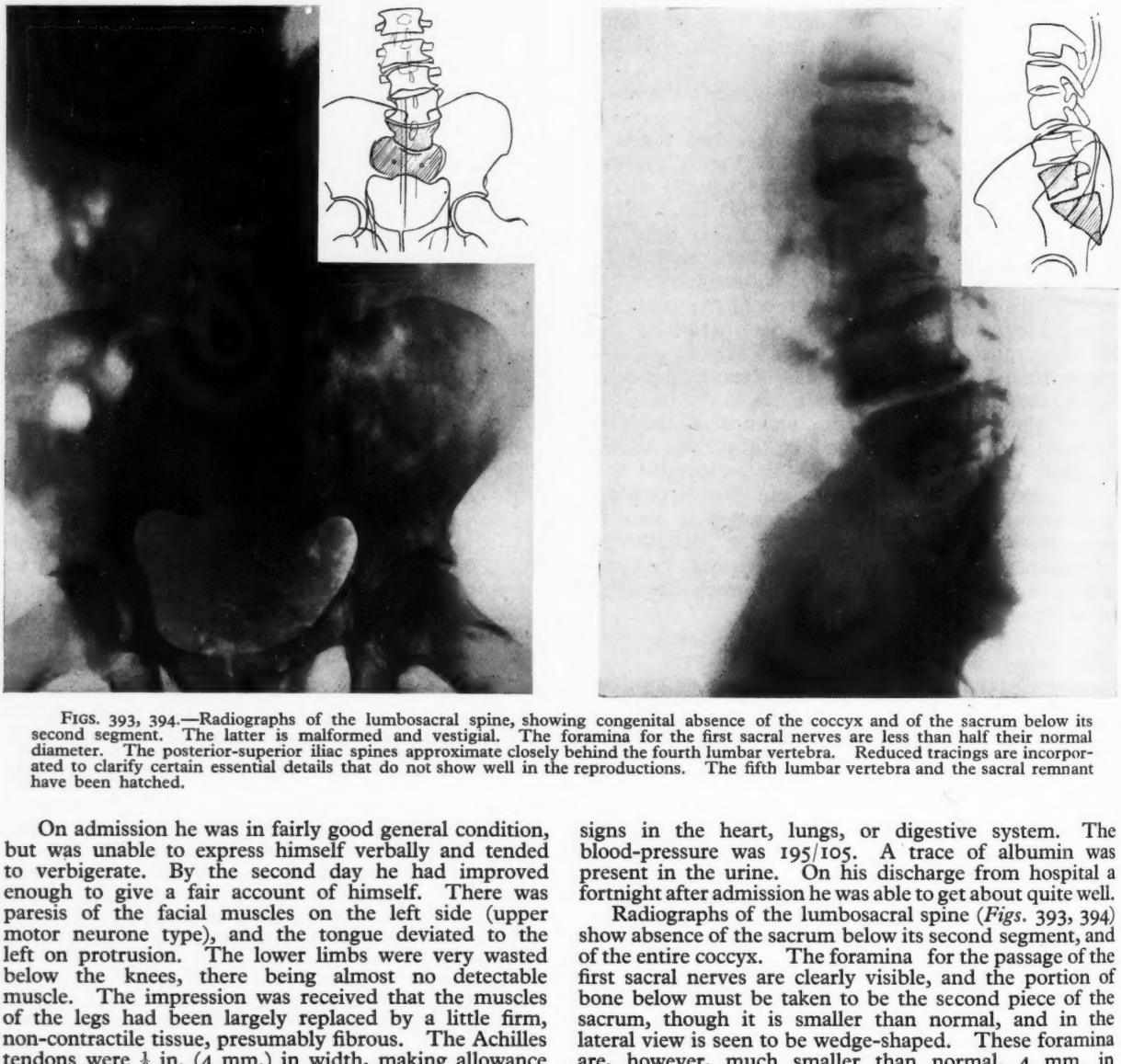
Treatment is (1) orthopædic, directed to the correction of deformities, and (2) genito-urinary, directed to the control of the urinary incontinence for which several operations have been devised (Aldridge, 1942; Price, 1933), and for which good results have been claimed.

Patients found to be suffering from congenital abnormalities of the rectum or urogenital tract should have their lumbosacral region X-rayed, since vertebral defects in this region do not necessarily cause symptoms.

CASE REPORT

The patient was a man aged 61, who had been resident for the previous seven years in an Institution. He was admitted to hospital because of the sudden onset of mental confusion with dysphasia.

He subsequently stated that his legs had been wasted ever since he could remember, certainly since earliest childhood, and presumably they had always been like that, but in spite of this he had been able to get about quite well. He had suffered from life-long urinary incontinence, and wore a portable urinal.



FIGS. 393, 394.—Radiographs of the lumbosacral spine, showing congenital absence of the coccyx and of the sacrum below its second segment. The latter is malformed and vestigial. The foramina for the first sacral nerves are less than half their normal diameter. The posterior-superior iliac spines approximate closely behind the fourth lumbar vertebra. Reduced tracings are incorporated to clarify certain essential details that do not show well in the reproductions. The fifth lumbar vertebra and the sacral remnant have been hatched.

On admission he was in fairly good general condition, but was unable to express himself verbally and tended to verbigerate. By the second day he had improved enough to give a fair account of himself. There was paresis of the facial muscles on the left side (upper motor neurone type), and the tongue deviated to the left on protrusion. The lower limbs were very wasted below the knees, there being almost no detectable muscle. The impression was received that the muscles of the legs had been largely replaced by a little firm, non-contractile tissue, presumably fibrous. The Achilles tendons were $\frac{1}{8}$ in. (4 mm.) in width, making allowance for skin thickness (normal = approx. $\frac{1}{2}$ in., 13 mm.). There was a flicker of voluntary movement, with some fibrillation, in the tibialis anticus muscles, but none was present in any other muscles that may have been present below the knee. Elsewhere the muscles showed merely the slight generalized wasting of a thin, old man. Sensation appeared to be normal, though it was not possible to test this accurately owing to inadequate co-operation resulting from his mental impairment. If any deficiency was present it must have been slight. The biceps, triceps, supinator, and knee-jerks were all present and equal. The ankle-jerks were absent. The abdominal reflexes were present. The right plantar response was extensor, the left was doubtful. The tone of the anal sphincter was extremely poor, and a ring of prolapsed mucosa with hæmorrhoids was present. The bladder was distended to within an inch of the umbilicus; there was some cicatricial phimosis due to recurrent sepsis resulting from the incontinence, and there was moderate benign enlargement of the prostate. The retention did not recur after catheterization. There was no deformity of the limbs or trunk other than that directly due to muscular wasting. There were no abnormal physical signs in the heart, lungs, or digestive system. The blood-pressure was 195/105. A trace of albumin was present in the urine. On his discharge from hospital a fortnight after admission he was able to get about quite well.

Radiographs of the lumbosacral spine (*Figs.* 393, 394) show absence of the sacrum below its second segment, and of the entire coccyx. The foramina for the passage of the first sacral nerves are clearly visible, and the portion of bone below must be taken to be the second piece of the sacrum, though it is smaller than normal, and in the lateral view is seen to be wedge-shaped. These foramina are, however, much smaller than normal, 4 mm. in diameter (normal = approx. 10 mm.), and presumably transmitted nerve-trunks of correspondingly diminished size. There was gross osteo-arthritis of the lumbosacral joint, and a fair amount in the remainder of the lumbar spine. The iliac bones approached each other closely behind a wedge-shaped fifth lumbar vertebra.

The condition necessitating his admission to hospital was a cerebral thrombosis, associated with arteriosclerosis and hypertension, with retention of urine due to moderate benign enlargement of the prostate and phimosis.

He was readmitted two months later in coma with a complete left-sided hemiplegia, and he died some days later from a terminal bronchopneumonia. Autopsy confirmed the presence of a thrombosis in the right middle cerebral artery, with softening of the infarcted part of the brain, and terminal bronchopneumonia. A carcinoma 2 in. (5 cm.) in diameter was present on

the greater curvature of the stomach, but there were no secondary deposits, and its presence does not appear to have contributed to his death. A large left-sided hydronephrosis and a prostate the seat of myo-adenomatous enlargement were present. The medical officer who performed the autopsy was apparently unaware of the congenital abnormalities and unfortunately the opportunity was not taken of obtaining a bony specimen, nor of dissecting the nerves and muscles.

SUMMARY

A man aged 61 was admitted to hospital suffering from a cerebral thrombosis. X-ray examination revealed absence of the sacrum below its second segment, and of the coccyx. There were gross deficiency of the musculature below the knees, life-long urinary incontinence, a lax anal sphincter, and hydronephrosis.

Congenital absence of the caudal end of the spine is rare, only 44 other cases having been reported during the past century.

In addition to the features described in this case report—namely, extreme wasting of all muscles below the knees, urinary incontinence, and a lax anal sphincter—which are typical of the condition, it is commonly associated with flattened buttocks, dislocation of the hip, talipes, and abnormalities of the pelvic viscera. The muscular wasting can be correlated with interference with or absence of the appropriate spinal nerve and vertebral segment. The previous literature is reviewed and a classification is presented in the table. The aetiology is discussed in terms of failure of an organizer or suppression of an evocator. Comparisons are drawn with similar conditions occurring hereditarily, sporadically, and experimentally in animals. The suggestion is made that the spine and pelvis should be X-rayed in all cases of congenital abnormality of the genito-urinary apparatus, rectum, or anus.

REFERENCES

ALBRECHT (1885), *Dtsch. med. Wschr.*, **11**, 413.
ALDRIDGE, A. H. (1942), *Amer. J. Obstet. Gynec.*, **44**, 398.
DE ARAÙJO, A. (1936), *Arch. brasil. Cir. Ortop.*, **4**, 43.
BARROS LIMA (1935), *Ibid.*, **3**, 195.
BERMAN, W. (1945), *Amer. J. Obstet. Gynec.*, **50**, 447.
BOWMAN, quoted by BRAILSFORD.
BRACK, E. (1929), *Virchows Arch.*, **272**, 295.
BRADBURN, W. R. (1922), *N. Orleans med. surg. J.*, **74**, 633.
BRAILSFORD, J. F. (1929), *Brit. J. Surg.*, **16**, 568.
CHARLIER, A., and SAUTY, P. (1913), *Rev. Orthopéd.*, **4**, 257.
DANFORTH, C. H. (1932), *Proc. Soc. exp. Biol., N.Y.*, **30**, 143.
DANKMEIJER, J. (1935), *Anat. Rec.*, **62**, 179.
DESFOSSES, P., and MOUCHET, A. (1924), *Rev. Orthopéd.*, **11**, 61.
DIAZ LIRA, E. (1938), *Rev. ortop. y traumatol.*, **7**, 231.
DREHMANN, G. (1927), *Beitr. klin. Chir.*, **139**, 191.
DUNN, L. C. (1925), *J. Hered.*, **16**, 127.
— — and LANDAUER, W. (1934), *J. Genet.*, **29**, 217.
— — — — (1936), *Ibid.*, **33**, 401.
ELMSLIE, R. C. (1921–2), *Proc. R. Soc. Med.* (Sect. Orthop.), **15**, pt. 3, 8.
ESPASA, H. S. (1928), *Rev. méd., Barcelona*, **9**, 547.
FELLER, A., and STERNBERG, H. (1931), *Virchows Arch.*, **280**, 649.
FITCH, R. R. (1910), *Amer. J. orthop. Surg.*, **7**, 540.
FOIX, C., ACHARD, C., and MOUZON, J. (1924), *Rev. Neurol.*, **31**, 270.
— — and HILLEMAND, P. (1924), *Ibid.*, 450.
FRIEDEL, G. (1910), *Arch. klin. Chir.*, **93**, 944.
GIRARD, P. M. (1935), *J. Bone Jt Surg.*, **17**, 1062.
GUILLEMINET, M. (1938), *Lyon chir.*, **35**, 369.
HAMSA, W. R. (1935), *Arch. Surg.*, **30**, 657.
HIGGS, S. L. (1924), *Proc. R. Soc. Med.* (Sect. Orthop.), **17**, 39.
HIGHMORE, NATHANIAL (1651), *History of Generation.*
HILGENREINER, H. (1937), *Z. Orthop.*, **66**, 224.
HOHL, A. F. (1852), *Zur Pathologie des Beckens*, 61.
KIENBÖCK, R., and ZIMMER, A. (1935), *Röntgenpraxis*, **7**, 111.
LAMOOT and GRAUX (1936), *Echo méd. Nord.*, **5**, 88.
LANDAUER, W. (1928), *J. Hered.*, **19**, 453.
— — (1933), *Ibid.*, **24**, 293.
LEHMANN, F. E. (1935), *Rev. suisse Zool.*, **42**, 405.
LERI, A., and LINOSSIER, A. (1925), *Bull. Soc. méd. Hôp. Paris*, **49**, 504.
LICHTER, A. (1947), *Arch. Surg.*, **54**, 430.
LITZMANN, C. C. T. (1884), *Arch. Gynaek.*, **25**, 31.
MALLY, M. (1903), *Bull. Soc. Anat.*, March, 322.
MÜLLER, J. H. (1936), *Röntgenpraxis*, **8**, 105.
NEEDHAM, J. (1942), *Biochemistry and Morphogenesis*, 370, 375. Cambridge.
NEWELL, J. L. (1934), *New Engl. med. J.*, **210**, 1217.
PÉREZ CASTRO, E. (1944), *Cir. App. locom.*, **1**, 104.
POUZET, F. (1938), *Lyon chir.*, **35**, 371.
PRICE, P. B. (1933), *Arch. Surg.*, **26**, 1043.
RENDU, A., and VERRIER, H. (1913), *Rev. Orthopéd.*, **4**, 311.
SINCLAIR, J. G., and DUREN, N. (1941), *Arch. Surg.*, **43**, 473.
STEWART, S. F. (1924), *Arch. Surg.*, **9**, 647.
WERTHEIM, C. C. (1857), *Mschr. Geburtsh. Frauenkr.*, **9**, 127.
WHITE, C. (1911), *Proc. R. Soc. Med.*, **4**, 279.
ZELIGS, I. M. (1940), *Arch. Surg.*, **41**, 1220.
ZILVA, S. S., DRUMMOND, J. C., and COWARD, K. H. (1921), *Biochem. J.*, **15**, 427.

EPIDERMOID CYSTS IN THE REGION OF THE RECTUM AND ANUS

A REPORT OF FOUR CASES

BY GEORGIANA M. BONSER, F. P. RAPER, AND H. S. SHUCKSMITH, LEEDS

THE surgical literature contains few reports of epidermoid cysts in the region of the rectum and anus, though there are many reports of epidermoid cysts associated with a pilonidal sinus. The cases reported here are most usually called presacral dermoids. Lockhart-Mummery (1914) describes a male patient with an anal fistula which terminated in a dermoid cyst anterior to the coccyx, and the same author (1934) includes in his book a report of a patient with a cyst lying between the rectum and sacrum which contained ciliated epithelium and clear fluid. Lahey and Eckerson (1934) reported 3 cases of presacral dermoids from the Lahey Clinic. Raven (1935), in a study of pathological specimens of sacrococcygeal cysts and teratomata, includes descriptions of three simple dermoid cysts, two

complex dermoid cysts, and a cyst thought to be derived from the post-anal gut. Whittaker and Pemberton (1938) report 9 cases of presacral dermoid

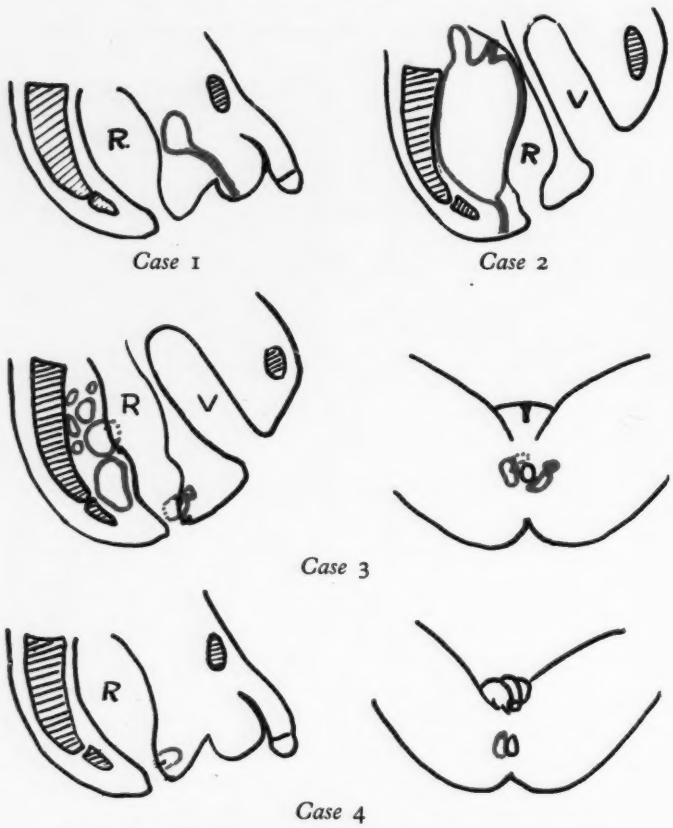


FIG. 395.—Diagrammatic representation of the site of the cysts in the four patients reported.

cysts seen at the Mayo Clinic between 1922 and 1936. Hayden (1939) states that he has seen several cases of fistulæ communicating with dermoid cysts

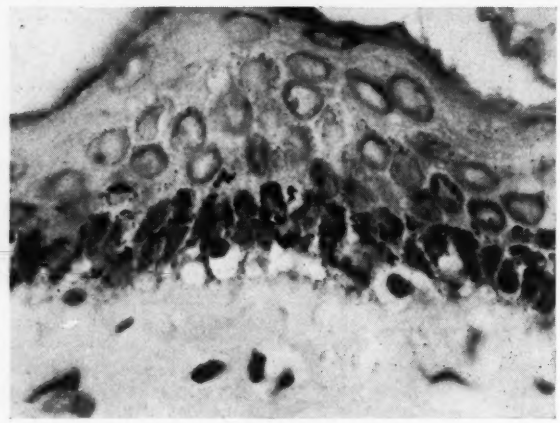


FIG. 396.—*Case* 1. Keratinizing, stratified, squamous epithelium lining the cyst. In the basal layers and to a lesser degree in the stratum granulosum are melanin granules. Note the tendency for the melanin to form a cap over the nucleus (well seen in the centre). Fontana. (× 600.)

in the retrorectal space; he describes one case in which there was a pilonidal cyst with several sinuses together with a presacral cyst quite unrelated to the sinuses. Manheim, Druckerman, and Peskin (1939) reported one case of presacral dermoid cyst seen at the Mt. Sinai Hospital and estimate that one such cyst occurred in 97,000 registrations at that hospital. Carney (1944) reports one case in which there was one large cyst and six to eight smaller cysts between the rectum and sacrum. One of the smaller cysts when examined was found to be lined by stratified epithelium.

CASE REPORTS

Case 1.—A boy aged 7 months had a small sinus in the midline at the lowest point of the scrotum (*Fig.* 395). It had been present since birth and small quantities of what was thought to be urine were passed from it occasionally. A sinogram showed a sinus 4 cm. long leading to a cyst the size of a hazel-nut, situated anterior to the rectum. At operation in August, 1948, the track and the cyst, which lay close to the rectum, were dissected out. There has been no recurrence.

On microscopical examination the wall of the cyst was found to be composed of vascular fibrous tissue lined by stratified squamous epithelium from 6 to 8 cells in depth and forming abundant keratin. In the cells of the basal layer is golden-brown melanin pigment (*Fig.* 396). No skin appendages are seen.

Case 2.—A girl aged 8 years had had since birth a sinus opening in the midline 2·5 cm. behind the anus (*Fig.* 395). It discharged a varying quantity of yellow, watery, or occasionally creamy fluid. On two occasions, when the sinus had remained closed for two to three weeks, there had been increasing difficulty in defæcation, which was immediately relieved when the sinus re-opened.

The sinus had been excised on five occasions (at 6 weeks, 1 year, 3 years, 5 years, and 6 years of age) but it had never healed. When seen in September, 1944, the sinus was emerging through scar tissue 2·5 cm. behind the anus, very slightly to the left of the midline. On rectal examination there was a large soft swelling lying between the sacrum and rectum, but only the lower half of the swelling could be felt. At operation in September, 1944, after removal of the coccyx, the cyst was excised. It was about the size of an orange and had two finger-like projections alongside the upper rectum. It contained several long hairs and thin sebaceous material with a very strong odour. There has been no recurrence of the sinus.

No histological examination was made.

Case 3.—A female, aged 28, of subnormal intelligence, complained of pain on defæcation of some weeks' duration. The mother stated that the daughter had had trouble with the seat as a baby; several abscesses had developed. Similar trouble had recurred in the early 'teens, but she had been free of symptoms for the last 13–14 years. Examination revealed the external opening of a fistula-in-ano 2·5 cm. from the anus in the left posterior position (*Fig.* 395). The rectum was loaded with fæces.

Three operations were performed in the last quarter of 1947. The fistula extended to the opposite side behind the anus and on the right a cavity containing pus was found lined with epithelium. Deep in the wound and behind the anorectal canal there was a large cystic tumour the size of a hen's egg and also a smaller cyst the size of a pea. At a second operation two cysts, one posteriorly and the other on the left, each the size

of a hen's egg, were removed, and on the third occasion, after excision of the coccyx, several more cysts about the size of grapes were cleared out from behind and from the sides of the rectum, extending as high as the sacral hollow.

Defection of the patient from hospital prevented a careful follow-up, but when seen fifteen months after the last operation rectal examination did not reveal any evidence of further cysts but the fistula was still present.

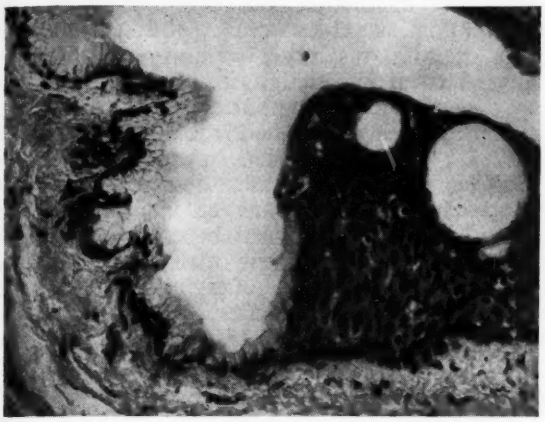


FIG. 397.—*Case* 3. Part of the lining of the smallest cyst removed at the first operation. On the left is a single layer of mucus-secreting epithelium; on the right is stratified epithelium many layers thick. H. and E. (× 140.)

From the first operation three pieces of tissue were examined—viz., the fistulous tract, a piece of the lining epithelium of the pus-filled cavity into which the tract opened, and the small pea-like cyst (0·5 cm. in diameter) which had been removed entire. Microscopically the tract is lined by dense inflammatory tissue, whereas the cavity into which it opens is lined by hyperplastic (up to 15 layers of cells) squamous epithelium forming keratin. The small cyst is lined by two types of epithelium: (*a*) a single or double layer of columnar cells secreting mucus; and (*b*) a stratified epithelium ranging from 2–15 cells in depth and without keratinization (*Fig.* 397). The transition from one type to the other is quite sharp; the extent of the stratified areas is greater than the columnar. In the deeper parts of the cyst wall are a number of small spaces lined by both types of epithelium and probably communicating with the interior of the cyst.

FIG. 399.—*Case* 3. Sebaceous glands and a hair follicle in the fibro-fatty tissue of the pre-sacral space removed at the third operation. H. and E. (× 40.)

At the second operation, the original pus-filled cavity was re-opened and a piece of the wall examined microscopically. As before, the lining epithelium consists of stratified squamous epithelium forming keratin. The epithelium is in part replaced by granulation tissue in which are clefts lined by foreign-body giant cells and foamy cells. In addition, two intact cysts with sebaceous contents and measuring approximately 1·5 in. in diameter were removed. No hairs were noted with the naked eye. These cysts proved also to be lined by keratinizing squamous epithelium or by granulation tissue where the epithelium had been shed. In the wall of one cyst are several groups of sweat-glands (*Fig.* 398).

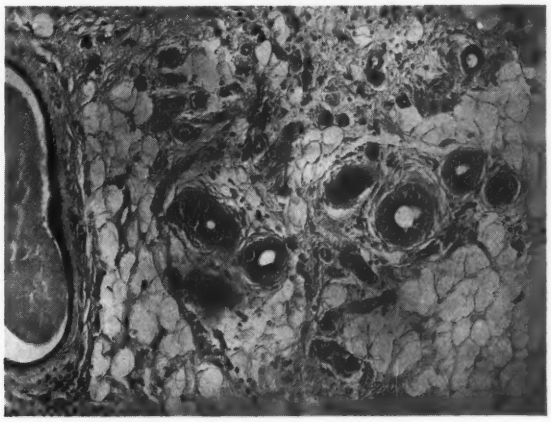


FIG. 398.—*Case* 3. A group of sweat glands in relation to the wall of one of the intact epidermoid cysts removed at the second operation. The cystic space on the left of the figure is a dilated sweat-duct. H. and E. (× 53.)

The fibro-fatty tissue removed at the third operation from the presacral space shows flakes of keratin which have induced a foreign-body giant-cell reaction and which are widely disseminated throughout the tissue. In addition, at a deep level, there are groups of sebaceous glands, one or two hair follicles, and some sweat-glands (*Fig.* 399).

FIG. 400.—*Case* 4. An island of ciliated epithelium, found in the lining of the cyst, of respiratory type flanked by stratified squamous epithelium. H. and E. (× 635.)

Case 4.—A male, aged 58, complained of pruritus ani and a swelling at the side of the anus present for one year. The irritation had existed for some months before the lump appeared. The patient thought that scratching had caused the lump and that it varied in size (*Fig.* 395).

On examination a subcutaneous cyst was found on the right anterolateral aspect of the anus; it was about

3 cm. in diameter and its centre was 2·5 cm. from the anal margin. It resembled a sebaceous cyst, though no central punctum could be demonstrated. In December, 1947, the cyst was excised completely from the superficial tissues and the area allowed to granulate. One year later there was no recurrence but the pruritus ani persisted and had led to chronic lichenification of the peri-anal skin.

The cyst is unilocular and measures 3 × 2 × 2 cm. The outer surface is partly covered by anal skin bearing a long hair. On section, turbid, pale-yellow fluid escaped and the inner lining was seen to be smooth and shiny. No hairs or other cutaneous appendages were noted. On microscopical examination, the cyst has a fibrous wall lined by somewhat compressed stratified epithelium from 3 to 5 cells in depth. For the most part this is undergoing keratinization but there is an island of ciliation and considerable numbers of mucus-secreting cells discharging into the cyst (*Fig.* 400). No skin appendages are seen.

DISCUSSION

Although the literature contains few reports of this type of cyst, the fact that we have seen four such cases within four years suggests that they may be more common than is realized.

Presenting Signs.—Three of our patients had fistulæ-in-ano. In the 21 cases which we have quoted from the literature, fistulæ had occurred in only 3 cases. The remainder had a cystic swelling between the rectum and sacrum which was discovered either on pelvic examination or which had become inflamed. One cyst had caused fatal intestinal obstruction (Raven, 1935). The site of the fistula in our *Case* 1 is unusual, but the site and structure of the cyst make it comparable with the other cases.

Age.—The ages of the cases previously reported together with our own cases range from two weeks to fifty-eight years. In 5 of the reported cases and in 3 of our own, the presenting symptoms arose in the first three years of life.

Sex.—The condition appears to be much more common in females than in males. In the reported cases in which the sex is known 15 were females and 4 males. In our own cases 2 were females and 2 males.

Curability.—We think it is important to recognize that these cysts may cause fistulæ-in-ano, especially in children, for it is impossible to cure such fistulæ unless the cyst is completely removed. It is also important to realize that the cysts may be multiple and that the tissue surrounding them (as in our *Case* 3) may contain elements capable of forming further cysts.

Nature of the Cysts.—The first specimen of this series to be examined microscopically was the small pea-like cyst removed entire from *Case* 3. The opinion was then expressed (G. M. B.) that as two types of epithelium were present, the cyst was probably of teratomatous nature. This view was much strengthened by the later findings in this case, when skin appendages such as sweat-glands (*Fig.* 398), sebaceous glands, and a hair follicle (*Fig.* 399) were seen. Additional support was gained from the presence of long hairs in *Case* 2, melanin-forming epithelium in *Cases* 1 and 3, and of ciliated epithelium in *Case* 4. Schwalbe (1907) accepted a distinction between bigerminal and monogerminal sacral teratomata, but thought that such a division was rather arbitrary and preferred to regard them as one group. He expressed the view that the more complicated the picture, the earlier in development had the aberration taken place. If this is accepted, then it should be concluded that in the cases described here, the developmental anomaly was of late origin. The tendency in recent years has been to discard rigid ideas about the development of the germinal layers and to regard all as capable of giving rise to a variety of different forms of epithelium. Thus the finding of mucus-secreting and ciliated epithelium of respiratory type together with epidermis and skin-appendages would not necessarily be an argument for the participation of two germinal layers in these structures. All might be derived from ectoderm and yet be regarded as teratomatous in nature.

SUMMARY

Four cases of epidermoid cysts in the region of the rectum and anus are described. Twenty-one similar cases have been collected from the literature. The condition is thought to be more common than is realized. Stress is laid (*a*) on the importance of recognizing such cysts as one cause of fistulæ-in-ano, especially in children; and (*b*) on the fact that the cysts may not only be multiple but the tissue surrounding them (as in our *Case* 3) may contain elements capable of forming further cysts. Reasons are given for regarding these cysts as teratomatous in nature.

REFERENCES

CARNEY, H. M. (1944), *Ann. Surg.*, **119**, 155.
HAYDEN, E. P. (1939), *The Rectum and Colon*, 331. Philadelphia: Lea & Febiger.
LAHEY, F. H., and ECKERSON, E. B. (1934), *Amer. J. Surg.*, **23**, 30.
LOCKHART-MUMMERY, P. (1914), *Diseases of the Rectum and Anus*, 187. London: Baillière, Tindall & Cox.
– – (1934), *Diseases of the Rectum and Colon*, 265. London: Baillière, Tindall & Cox.
MANHEIM, S. D., DRUCKERMAN, L. J., and PESKIN, H. (1939), *Mt Sinai Hosp. J.*, **6**, 31.
RAVEN, R. W. (1935), *Brit. J. Surg.*, **23**, 337.
SCHWALBE, E. (1907), *Die Morphologie der Missbildungen des Menschen und der Tiere*, **2**, 361. Jena: Gustav Fischer.
WHITTAKER, C. D., and PEMBERTON, J. DE J. (1938), *Ann. Surg.*, **107**, 96.

HÆMOSTASIS WITH ABSORBABLE ALGINATES IN NEUROSURGICAL PRACTICE

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THE essential part of neurosurgical technique is prompt and lasting hæmostasis. This is for the most part accomplished by diathermy coagulation, but in certain circumstances this method is not applicable, as, for example, in the case of bleeding from venous sinuses, bone, and in some instances from extradural veins.

The materials used for hæmostasis are Horsley's wax for osseous bleeding, and fibrin† foam with thrombin (Bailey and others, 1946), oxidized cellulose, with or without thrombin (Frantz, 1945), or gelatin foam with thrombin (Light and Prentice, 1945) for the other types. The last three substances mentioned belong to the group of absorbable hæmostatics developed in the United States during recent years.

Recently one of us (G. B., 1946, 1947 *a*, *b*) has reported on absorbable alginate materials. Their uses in other branches of surgery have also been described (Passe and Blaine, 1948; Mullard, 1948). Alginates appear to have important advantages over the other absorbable hæmostatics so far developed. They are heat sterilizable, and are available in a number of physical forms and various grades of solubility and absorbability. Alginates have been developed entirely in Britain, whilst all other modern absorbable hæmostatics have been developed in the United States.

Like oxidized cellulose, alginic acid is a polyuronic acid. As such it is a member of a large number of naturally occurring substances extractable from vegetable matter. It is found in the extract from brown seaweeds growing off the Scottish and Irish coasts and is being recognized more and more as a nationally important raw material having many uses in an increasing number of industrial and scientific applications, for which, hitherto, imported raw materials had to be used.

The parent extraction product is alginic acid (a polymer of mannuronic acid), the empirical formula of which is $C_6H_{10}O_7$ with an equivalent molecular weight of 194. The carboxyl groups of this acid react with metallic or ammonium ions forming corresponding alginates, some of which, like the sodium salt, are water-soluble, possessing marked hydrophilic colloidal properties, whilst others, such as the calcium salt, precipitate an insoluble alginate. Thus, for instance, the interaction of sodium alginate with a calcium salt produces an instant coagulum, a reaction which makes it possible to produce in the industrial laboratory a wide range of physical products such as films, yarns, gauze, wool, and foams.

Previous work has shown that calcium alginate materials are slowly absorbed when implanted into living tissue, the rate of absorption depending on the amount used and the vascularity or reactivity of the tissue bed.

Further work (being published elsewhere) has shown that the absorption rate of these materials can be easily altered by a method of increasing the sodium content of the molecular structure. Unlike oxidized cellulose, which belongs to the same group of materials, alginates, by virtue of their stability at physiological levels of hydrogen-ion concentration, do not retard bone repair, nor do they interfere with penicillin or thrombin activity; furthermore they possess the practical advantage of standing up to sterilization by autoclaving or by dry heat.

The specific hæmostatic properties of absorbable hæmostatics other than fibrin foam and thrombin must be regarded with reserve. Frantz and coworkers have postulated a specific hæmostatic action of oxidized cellulose which seems to depend on the formation of a coagulum consisting of salts of polyanhydroglycuronic acid and hæmoglobin. No similar claim is made with reference to alginates beyond the obvious mechanical hæmostatic action inherent in any material which by its physical structure as a foam, wool, or gauze, provides a greatly increased clotting surface. The possible specific hæmostatic properties of alginates are, however, being investigated elsewhere.

In animal experiments calcium alginate is usually absorbed within three months. In highly vascular beds absorption is rapid, whilst if the material is inserted into an avascular tissue plane, it is greatly delayed. In some cases when material is inadvertently treated with antiseptics or antifungicides (e.g., C.T.B.)‡ during its production absorption is not completed. Such 'impure' alginate provokes a copious serous exudate after a few days, with a concomitant swelling of the material; on removal the material always looks like 'seaweed jelly',

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† We are indebted to Professor Norman M. Dott, Edinburgh, for the following correction relating to the use of fibrinogen or fibrin: Mr. Dickson Wright was the first to suggest its use in 1940 at Oxford, at a meeting of the Society of British Neurological Surgeons. Professor Ian Aird mentioned this in an Annotation on Fibrin in Surgery in the *British Medical Journal* after the war.

‡ C.T.B. is cetyl pyridinium bromide (Fixanol I.C.I.) which is bactericidal to staphylococci at a concentration of 1 : 1,000,000. In some samples the concentration of this substance was approximately 1 : 500. When reduced to a concentration of 1 : 100,000 it provoked no marked reaction.

undergoing some degree of marginal digestion. On histological preparations of such specimens at later stages after operation, signs of a chronic inflammatory process are seen, with fragmented remnants of the alginate fabric encapsulated by fibrous granulation tissue studded with giant cells. Material not so treated (' pure calcium alginate ') produces no encapsulation or giant-cell reaction.

FIG. 401.—Photomicrograph of trephine hole (3 mm.) in skull of rabbit 65 days after operation, at which a plug of ' slow ' alginate wool was inserted into the bone gap. The gap is firmly bridged with fibrous tissue with the fibres of the neomembrane directed horizontally. There is no macroscopical evidence of alginate wool. (× 6·5.)

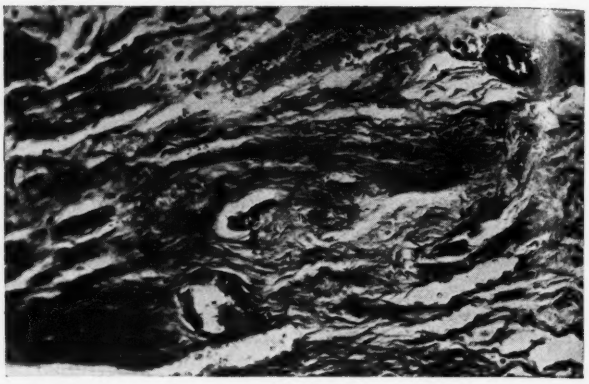

FIG. 402.—Higher magnification of part of *Fig.* 401. A few small fragments of alginate wool are seen in process of organization. The fibrous tissue presents a healthy appearance with a marked absence of a foreign-body reaction around the last remnants of the alginate wool. The fragments are in course of final digestion by macrophages. No giant cells are visible. (× 145.)

Alginate materials with a higher sodium content are absorbed and become soluble very much more quickly, i.e., in about two weeks or less. Under aseptic conditions the tissue reaction is usually an aseptic inflammation, as with the other absorbable hæmostatics: first there is a polymorphonuclear leucocytic response which subsides in a few days, giving place to infiltration of round cells and of lymphocytes, which in turn are replaced by a moderate degree of fibrosis and macrophages. After final digestion of the alginates only a minimal scar is found on microscopical examination.

Figs. 401, 402 shows the state of affairs in the dura of a rabbit 65 days after operation, at which time only a minute residue of microscopical algin fragments is demonstrable in an otherwise healthy dura over normal brain tissue; the animal had no focal symptoms of any kind during its experimental life.

Figs. 403–405 shows the appearances of scar tissue in the anterior abdominal wall of a rat seven days after insertion of quickly absorbable alginate wool into the abdominal cavity. Approximately 1 g. of wool was inserted and the specimen represents the only area where alginate remnants were detected.

At the Neurosurgical Centre, Oldchurch Hospital, Romford, the use of alginates as absorbable hæmostatics has now been the routine practice for 18 months and it is timely therefore to report our observations.

RESULTS

In this clinical trial alginate wool and an alginate bone-wax substitute were used.

1. Wool.—Early in our experience it was found that alginate wool was the most suitable physical form for neurosurgical work because it allows the use of suction apparatus whether the wool be used dry or wrung out in saline. We have used alginate wool only in those cases where the nature of the hæmorrhage made it inexpedient or dangerous to use the coagulating current. In all cases we were impressed by the rapid, almost instantaneous, hæmostatic effect produced.

The following table shows the type and number of cases in which alginate wool was used:—

Titles	*Number of Cases*
Laminectomy	19
Posterior fossa exploration	11
Plastic operation on the skull	3
Needling of brain	9
Removal of brain tumour	4
Operation for meningocele	1
Division of fifth-nerve roots	6
Removal of acoustic tumour	1
Cordotomy	13
Operation for hydrocephalus	1
Anterior fossa exploration	16
Total	84

The following case records illustrate the use of the material:—

Case 1.—A. M., male, aged 42. Diagnosis: Pineal tumour. Operation Jan. 8, 1948 (L. C. O.): A bone-flap was turned in the right parietal region. The dura mater was then reflected towards the superior longitudinal sinus and during this procedure an abnormally large cortical vein was damaged at its entrance to the sinus, causing an alarming hæmorrhage. Prompt application of alginate wool stemmed the deluge. The wool was left in place and the flap was closed. Apart from transient diplopia on the second day and a small rise of temperature on the fourth post-operative day, the patient made an uneventful recovery. No signs or symptoms attributable to a foreign body were found at periodic follow-up examinations to date.

Case 2.—M. S., female, aged 20. Clinically diagnosed as ? posterior fossa tumour. Ventriculography showed displacement of ventricles downwards and to the left. Operation (Jan. 13, 1948) (L. C. O.): Right frontal flap was turned and, after needling, an encapsulated, lobulated tumour 1½ in. in diameter was removed. The resulting hæmorrhage was controlled by packing

the cavity with alginate wool. Subtemporal decompression was carried out, the dura was left open, and the wound closed. Histology of smears had previously shown the tumour to be of a malignant and undifferentiated type.

In spite of good recovery from the operation the patient suddenly died 12 hours after the operation. At autopsy the tumour cavity was seen to be filled with alginate wool which formed a jellified mass of soft consistency and a brownish-red colour. There was no sign of protracted hæmorrhage. The wool showed considerable marginal liquefaction.

Case 3.—J. W., male, aged 29. Diagnosis: Head injury (road accident). The patient was able to localize his injury to the right parietal area. After clinical

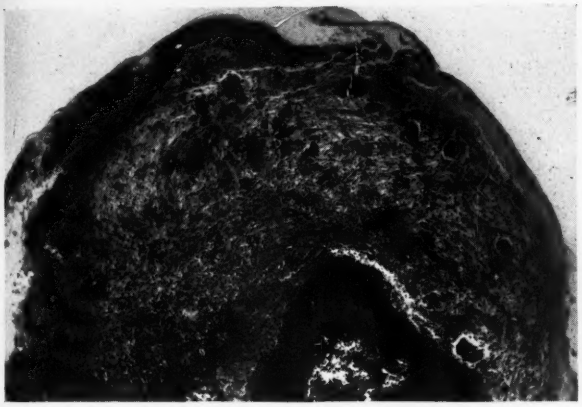

FIG. 403.—Photomicrograph of experimental wound in anterior abdominal wall of a rat at seven days post-operatively. At operation 'fast' alginate wool was inserted into the abdominal cavity. The section shows part of the parietal peritoneum with numerous alginate wool fragments embedded in scar tissue. There were no other sites of 'embedded alginate' in the animal. (× 50.)

examination a diagnosis of left middle meningeal hæmorrhage was made and in view of the rapid deterioration in the patient's condition during his short observation in hospital exploration was decided on.

Operation (Jan. 13, 1949) (J. W. M.): A large burr hole was made over the left hemisphere and, as soon as the bone was perforated, dark blood-clot was forced out through the opening. On enlarging the burr hole with bone-nibbling forceps a thick, fairly firm clot was seen overlying the dura and extending upwards, backwards, and downwards. The extension of the opening was continued in those directions and the clot was removed by irrigation and suction as far as possible.

The clot was found to extend into the occipital area, and upwards it reached the superior sagittal sinus. Exploration of the branches of the middle meningeal artery showed the artery to be intact except for a very small leak from its anterior branch; the origin of massive bleeding was a tear in the superior longitudinal sinus from which a good deal of blood was flowing.

After evacuation of the clot alginate wool was packed up towards the sinus, arresting the hæmorrhage. This area controlled, there was no further hæmorrhage from the dura or from the subdural space, and after insertion of a drain extradurally the wound was closed and the alginate wool left in situ.

The patient's recovery was uneventful and there were no focal signs or symptoms on his discharge from hospital twenty days after the operation. Follow-up examination three weeks later found the patient in good condition, with no residual symptoms.

This case was particularly noteworthy as there was some traumatic bruising of the patient's cerebral cortex and he had, correspondingly, a paresis of the right arm and leg for a few days post-operatively. This paresis cleared up completely during his convalescence and no neurological sign or symptom was found which could be attributed to an unabsorbed foreign body pressing on his cerebrum.

Case 4.—H. B., female, aged 59. Clinically diagnosed as a tumour of the pituitary region. Operation (June 2, 1948) (L. C. O.): Left frontal burr hole was

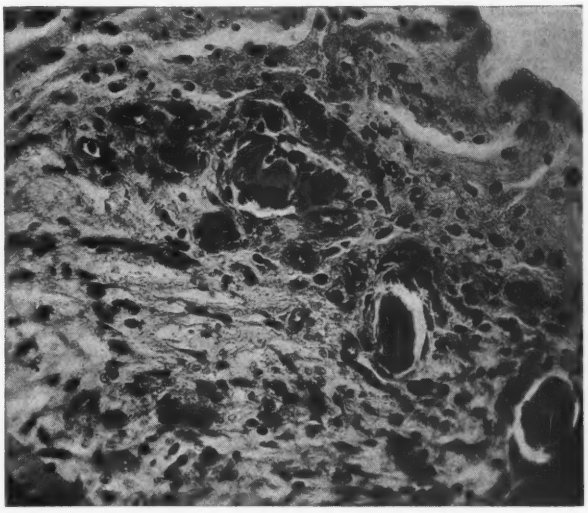

FIG. 404.—Higher magnification of a portion of the preceding section. Alginate wool fragments are seen in process of absorption by round cells and macrophages. A few polymorphonuclears are still present. There are a few giant cells; fibrous repair is well advanced. There is no marked foreign-body reaction around the alginate fragments. (× 205.)

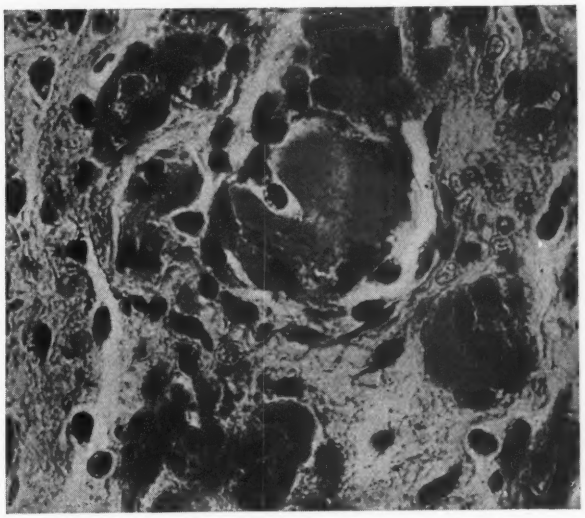

FIG. 405.—Photomicrograph of an alginate wool fragment from the preceding section. The picture shows the digestion of the absorbable wool by round cells and macrophages. There are no inflammatory phenomena to be seen. (× 580.)

made and the frontal lobe needled. No tumour was found on the left side. Needling was repeated through right frontal burr hole and a resistance was encountered at a depth of 4½ cm. Smears were made of the material aspirated and a biopsy taken with a punch. Considerable hæmorrhage resulted, which was adequately controlled with alginate wool.

The pathological report on the smears showed the tumour to be a glioblastoma multiforme.

The patient died on July 28, and an autopsy was undertaken. The following is an extract from the autopsy report :—

Meninges N.A.D.

Cerebrum : right larger than left. A yellowish tumour hard in patches present in right frontal lobe 2 in. across, diffuse, and invading the surrounding brain. The rest of the right cerebral hemisphere œdematous. Ventricles dilated. Cerebellum and pons N.A.D. No sign was found of the alginate wool inserted at operation, and there were no adhesions of the meninges or reactive signs at the site of insertion.

Histology confirmed the diagnosis of glioblastoma multiforme.

2. Bone Wax Substitute.—It is known that Horsley's wax produces a foreign-body reaction. One of us (L. C. O.) recently had experience of a case demonstrating this effect :—

I. D., female, aged 40. Three-and-a-half years ago operation at another hospital for right-sided middle meningeal hæmorrhage, from which the patient made an uneventful recovery. Two years later she developed two sinuses on the right side of the head, discharging pus. These failed to respond to chemotherapy. Operation (Jan. 28, 1947) : A small scalp-flap was turned down and granulation tissue appeared through the original bony opening. Exploration of the latter revealed a quantity of caseous material which was removed and subsequently shown to be bone-wax. The wound healed quickly after operation. The patient, when seen again in April, 1948, had experienced no further trouble.

Similar cases will probably be more frequently reported in the future under the stimulus of the recent number of papers on talcum-powder granuloma in general surgery (Eiseman and others, 1947 ; Mackey, 1948).

The rationale of the use of an alginate product as a bone-wax substitute was made possible by the versatility of this chemical, making it possible to produce material which, when in contact with a moist surface, undergoes an instant controlled swelling and conversion into a soft, waxy, homogeneous jelly. We are also told by the makers that it will eventually be possible to produce a wax-like jelly from this material.

The alginate ' bone-wax substitute ' used by us was borated alginate wool with a high sodium content which we made up into small pellets and subsequently rubbed into bone with a dry forceps or dry gauze. This latter precaution is necessary because the material naturally adheres to a moist instrument or glove. Instant hæmostasis was achieved by this means on all occasions.

It is worthy of note that some grades of alginate wool experimented with in the early stages of the adaptation of the wool as a bone-wax substitute were too soluble : the material was seen to disintegrate and absorb in under a quarter of an hour. In later trials we therefore used alginate wool of a more delayed solution rate, i.e., of a lower sodium content.

DISCUSSION

We feel that alginate wool has many advantages over its contemporaries by virtue of heat resistance and non-interference with the action of antibiotics and thrombin. The versatility of the material makes it possible to adapt it to the varying needs of surgery. ' Pure ' calcium alginate wool has the drawback of slow absorbability, which is mainly apparent in sparsely vascularized sites. This slow absorption is to be expected with an insoluble calcium salt, and is the recognized process in the organization of ' homogeneous ' absorbable materials like a fibrinous exudate or, even more so, of bone.

Slow absorption in poorly vascularized sites has also been noted when fibrin, gelatin foam, and oxdized cellulose are used. This response might be explained by noting the fact that fibrin and gelatin foams are formalin treated to facilitate their production, whilst oxidized cellulose, at a *p*H of under 4·0, has oxidization by-products. Thus an irritant constituent might well give rise to a fibrous reaction or interfere with macrophage activity.

It is therefore a welcome fact that such a highly respected ion as sodium is the factor influencing the absorption and solution rate of alginates, and that the chemistry of alginates allows full use to be made of this salt without destroying the essential physical properties of the material.

SUMMARY

1. Alginate wool has been found to be hæmostatic and absorbable at varying rates in animal experiments.

2. Alginate materials have been found to be non-toxic in animal experiments and have proved successful on clinical trial.

3. A case has been made out for the use of alginate wool as an absorbable hæmostatic in neurosurgical practice.

4. Attention has been drawn to its properties, physical forms, alterable solution rates, and general versatility, which should render alginate an important material in modern surgical technique.

We acknowledge with thanks the help given us by the Scottish Seaweed Research Association and Mr. C. W. Bonniksen, B.Sc., on the supply and chemistry of alginates. We also thank Dr. A. Glucksman, Strangeways Research Laboratory, Cambridge, for advice on histology ; Mr. E. Winter, Strangeways Research Laboratory, for preparing the sections ; and Mr. E. Wilmott, British Postgraduate Medical School, for the excellent photomicrographs. We also thank the chemists and research personnel of Messrs. Courtaulds Ltd., and Perivale Laboratories Ltd., for the many modifications to the material carried out at our request.

REFERENCES

BAILEY, O. T., et al. (1945), *Surgery*, **18**, 347.
BLAINE, G. (1946), *Lancet*, **2**, 525.
— — (1947, a), *Ann. Surg.*, **125**, 102.
— — (1947, b), *Med. Pr.*, **218**, 5650.
EISEMAN, B., and others (1947), *Ann. Surg.*, **126**, 820.
FRANTZ, V. K. (1945), *Surg. Clin. N. Amer.*, **25**, 338.
LIGHT, R. U., and PRENTICE, H. S. (1945), *Arch. Surg.*, **51**, 69.
MACKEY, A., and others (1948), *Brit. med. J.*, **1**, 1090.
MULLARD, K. S. (1948), *Thorax*, **4**, **3**, 233.
PASSE, E. R. GARNETT, and BLAINE, G. (1948), *Lancet*, **2**, 651.

CARCINOMA OF THE ADRENAL CORTEX

REPORT OF A CASE WITHOUT ENDOCRINE CHANGES

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TUMOURS of the adrenal gland are classified according to whether they arise from the medulla or the cortex. Those arising from the cortex are uncommon. A very high proportion are associated with endocrine changes. This is seen from recent reports (Kepler and Kealing, 1941 ; Cahill, Melicow, and Darby, 1942).

It is clear that these endocrine changes have been overlooked by early authors, who relied largely on post-mortem material (Kelynach, 1897 ; Rolleston and Marks, 1898). Kelynach (1897), reviewing cases of adrenal tumours collected from the earlier literature, claims that many of the reported adrenal carcinomata are manifestly secondary, and that the majority of reports are incomplete.

The demarcation between adenomata and carcinomata in the adrenal cortex is ill-defined. Willis (1948) considers it to be a matter of personal preference whether a large circumscribed but growing tumour shall be regarded as an unusually active adenoma or a carcinoma which has not yet displayed invasiveness or metastases.

This report concerns a woman with a carcinoma of the adrenal cortex but no endocrine changes. The case is also of interest in that the tumour was extensively calcified, as demonstrated radiographically.

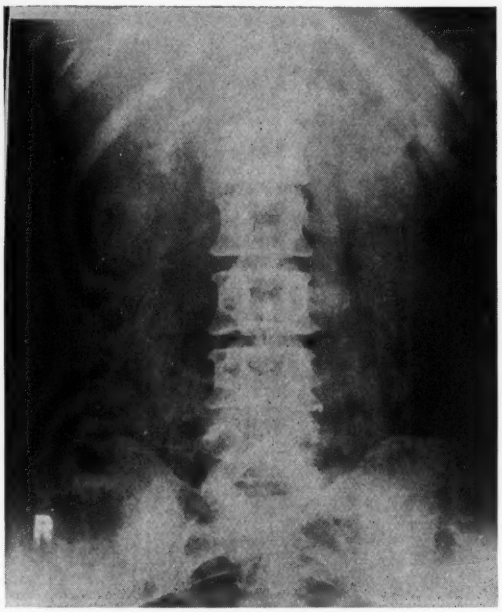

FIG. 406.—A plain radiograph of the abdomen in December, 1947, showing a collection of opaque material in the right hypochondrium in the region of the tip of the 12th rib.

FIG. 407.—An intravenous pyelogram in December, 1947, showing the relationship of the right kidney to the opaque shadow, and slight dilatation of the calices and pelvis on the right side.

CASE REPORTS

Mrs. N. G., a housewife aged 59, first reported with epigastric pain in April, 1947. She was relieved by a short course of dietetic treatment.

In February, 1947, she reported to the gynæcological department following a sudden loss of blood per vaginam. Palpation of the abdomen at that time showed no abnormality. A pre-operative diagnosis of carcinoma of the uterus was made.

In the sexual history there was no suggestion of general endocrine changes. The menses had begun at the age of 11 years and had always been regular and normal. She had four pregnancies with live children and no miscarriages. The menopause had occurred at the age of 49.

She was admitted to hospital and a pan-hysterectomy was performed. She was found to have a fibromyoma of the uterus.

In April, 1947, she again attended with severe abdominal pain starting in the region of the umbilicus, passing up to the epigastrium and then through to the back. She had vomited on occasions and her appetite had been poor. On examination she was tender in the epigastrium and right hypochondrium. A barium meal showed a normal gastric outline, but a fairly marked duodenal deformity with evidence of an ulcer. The abdominal pain was relieved by diet and alkalis. In September, 1947, she was well apart from occasional pain, and continued on a peptic ulcer régime.

In December, 1947, she developed sudden, severe, colicky pain starting in the right lumbar region, and radiating to the right loin, right groin, and occasionally into the right thigh. It lasted for about three hours. There was tenderness and guarding in the right hypochondrium and right posterior renal angle. This attack was thought to be renal colic.

A plain radiograph of the abdomen showed a curious collection of opaque material in the right hypochondrium in the region of the tip of the 12th rib (*Fig.* 406).

Cholecystography was normal. An intravenous pyelogram showed only slight dilatation of the calices and pelvis on the right side. The relationship of the right kidney to the opaque shadow is seen in *Fig.* 407.

Examination of the urine showed no abnormality. The examination of the stools for occult blood was negative on three occasions.

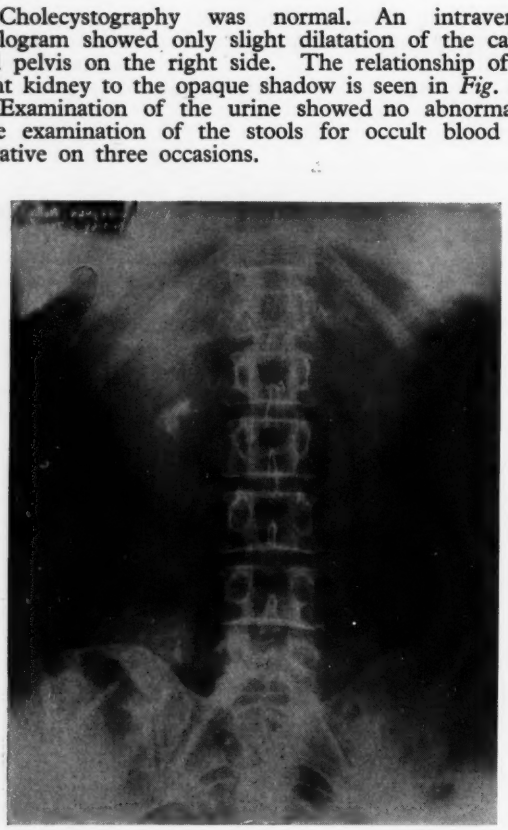

FIG. 408.—A plain radiograph of the abdomen in June, 1948, showing the opaque calcareous material in the right upper abdomen at a lower level than in the previous film.

She was again admitted to hospital in June, 1948, with severe pain of six hours' duration. A provisional diagnosis of renal colic was made. She had experienced three such attacks, since her previous admission, lasting up to three days and sometimes associated with nausea. In between these attacks she had a persistent dull ache in the right side. Her appetite was poor, she was often nauseated, she complained of malaise, and thought she had lost weight.

ON EXAMINATION.—She was tender on the right side, and the right kidney could now be felt without difficulty.

The findings on special investigation were :—

There was a slight secondary anæmia (Hb 66 per cent, R.B.C. 2,900,000 per c.mm.).

The alcohol test-meal was normal, and there was no occult blood in the stools.

A plain radiograph of the abdomen showed as before the presence of opaque calcareous material in the right upper abdomen, but it lay at a lower level than in the previous film. (*Fig.* 408.)

The barium meal was repeated and showed what was believed to be an ulcer on the posterior duodenal wall.

The examination of the urine was normal, and an intravenous pyelogram showed as before a slight dilatation of the calices and pelvis of the right kidney and a marked downward displacement of the organ. (*Fig.* 409.)

AT OPERATION.—On July 1, 1948, an operation was performed by Mr. F. F. Rundle through a right oblique lumbar incision. The tumour was densely adherent to the upper pole of the right kidney and seemed, in fact, to be in it; the kidney was therefore removed with the neoplasm, as was the overlying peritoneum.

Convalescence was uneventful.

Seventeen-ketosteroid estimation in the urine immediately after operation was 5 mg. per cent.

PATHOLOGICAL REPORT.—The specimen removed consisted of a kidney normal to external appearance together with a mass of tumour tissue measuring 10 by 8 by 6 cm. and separated from the upper pole of the kidney by connective tissue (*Fig.* 410).

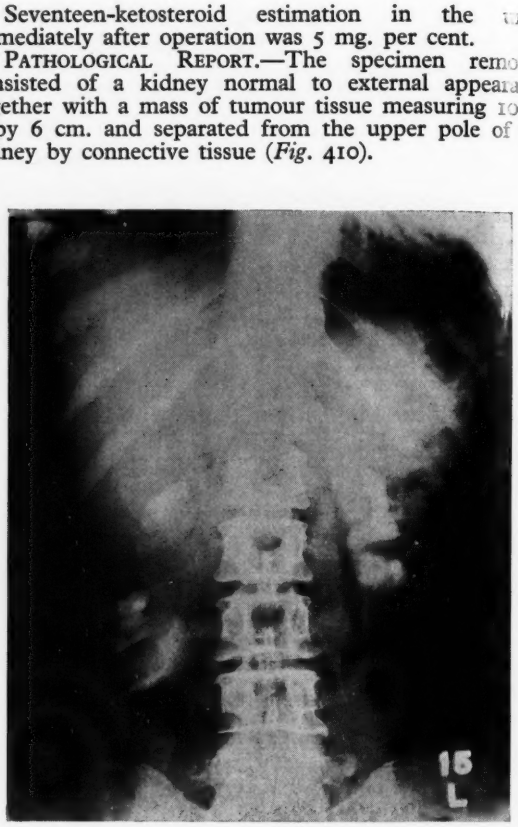

FIG. 409.—An intravenous pyelogram in June, 1948, showing marked downward displacement of the right kidney and dilatation of the calices and pelvis.

FIG. 410.—Cut section through tumour and kidney. (×⅓.)

The tumour was coarsely lobulated on the surface. Cut section showed the tumour to be divided into two unequal portions. The septum dividing those portions consisted of fibrous tissue together with a yellowish-coloured strand tissue which resembled the normal suprarenal cortex, and also contained a calcified area measuring 2 by 1 cm. The smaller portion was composed of necrotic tumour material and showed recent hæmorrhage. The larger portion was irregularly divided into lobules by strands of fibrous tissue passing in from the capsule. The appearance of these lobules varied from white tumour tissue to necrotic areas containing old and recent hæmorrhages (*Fig.* 410).

Histological Report.—The neoplasm was composed of fairly large eosinophil polyhedral cells which were lightly packed or grouped together in columns and separated by thin-walled sinusoids resembling an 'endocrine' pattern. In some areas poorly formed 'acini' were seen, and occasionally the stroma had degenerated and the cells had taken on a spheroidal appearance. Occasionally small necroses were present. Mitoses were fairly frequent and Scharlach R stain revealed a little fat content. Vine's stain in frozen section appeared negative.

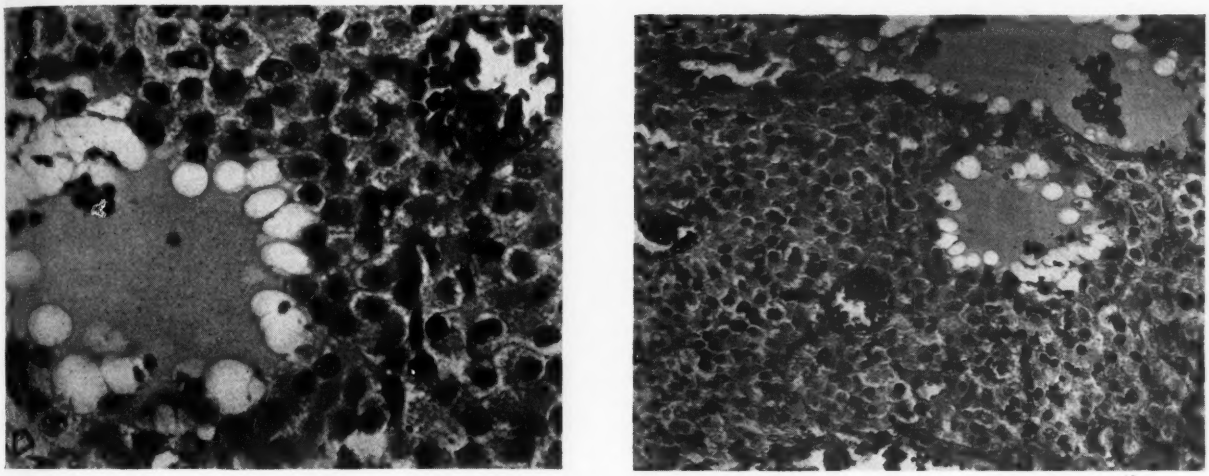

FIG. 411.—Microphotographs of a section of the tumour (*see* text).

The histological appearances show that this was a carcinoma of the adrenal cortex, although there was no definite evidence of invasive properties. *Fig.* 411 shows microphotographs of a section.

The patient has been followed up after discharge. A barium meal in September, 1948, showed a normal stomach and duodenum. She is at present in good health—her weight is keeping constant—and there is no evidence of recurrence of the neoplasm.

Comment.—The patient had attended for nearly eighteen months with infrequent attacks of severe upper abdominal pain. She had been variously diagnosed as suffering from peptic ulcer and biliary and renal colic (Dietl's crises). Eventually the right kidney was displaced downwards. In retrospect it is clear that the calcified area indicated a tumour displacing the kidney downwards, and this suggestion was made by Dr. G. Simon when he reported on the radiographs.

DISCUSSION

Wu (1940), in an exhaustive search of the literature, found only 4 cases in females of adrenal cortical carcinoma without endocrine changes. He claims that adrenal cortical carcinoma fails to produce changes only after the menopause. He describes female patients aged 62, 64, and 66 years respectively who did not show endocrine changes, and casts considerable doubt on the lack of these changes in the reported case of a female aged 36. Cahill, Melicow, and Darby (1942) have reported on 2 female patients with adrenal cortical carcinoma without endocrine changes, one occurring before the menopause at the age of 38, and another at the age of 50, after the menopause. Loeb (1941) also reported a case in a female aged 48, whose periods ceased at the age of 40.

The symptomatology in the case here recorded falls into line with the previously reported cases of this kind. Due to its deep situation, such a tumour without endocrine changes may initially be without symptoms. The condition causes pain when it is large enough to exert pressure on the kidney and other adjacent structures. Thus the tumour may be of large size when the patient is first seen. The displacement of the stomach as described by Shentone and Lougheed (1936), and by Alexander (1944), and the distortion of the duodenum, as in this case, may cause symptoms of gastro-intestinal disorder and give rise to a filling defect on barium-meal examination.

In this case, and in the cases described by Hermann (1944) and others, the kidney showed evidence of downward displacement. This renal displacement may result in a patient presenting with symptoms suggestive of renal colic or Dietl's crisis, due to pelvi-ureteric obstruction. This displacement can be demonstrated first by urographic investigation, as discussed in a recent paper by Kaplan and Greene (1948); and, eventually, on palpation when the neoplasm has reached a large enough size.

A history of malaise, loss of weight, and a palpable abdominal mass form a common triad of symptoms in these cases when the patient is first seen. This was emphasized by Loeb (1941). Using perirenal air insufflation he made a correct preoperative diagnosis of adrenal neoplasm in a patient without endocrine changes.

Calcification occurring in carcinomata of the adrenal cortex is not mentioned in standard works on radiography. Samuel (1948) discussed the origin

of calcification in the right hypochondrium, and describes a case of calcification in a carcinoma of the adrenal cortex associated with adrenal virilism. The present case report is an example of calcification in an adrenal cortical neoplasm with no endocrine changes.

The high operative mortality associated with the removal of adrenal cortical tumours has been attributed to various causes by different authors. Cecil (1933) states that in 39 per cent of adrenal cortical tumours subjected to surgery, the patients die of shock. This shock he says is due to the toxic effects produced by the expressing of large amounts of secretion into the blood-stream. Cahill (1942) attributes the high operative mortality to acute adrenal cortical deficiency. Cecil and others (1933) claim that in 30 per cent of cases with adrenal cortical tumours the opposite suprarenal is absent owing to atrophy. Lukens, Flippen, and Thigpen (1937) report examples of congenital absence of the adrenal gland on one side. Acute cortical deficiency has not been reported after the removal of a cortical carcinoma without endocrine changes, but Cleveland and Knox (1943) describe a case of bilateral carcinoma of the cortex with metastases in which acute cortical deficiency did develop following a relatively simple surgical exploration.

In discussing operative procedure Cecil (1933) makes the point that the suprarenal vessels, especially the veins, should be tied before manipulating the tumours, so as to prevent the squeezing of any secretion into the general circulation. He recommends a transperitoneal approach to the tumour so that this can be done with the greatest ease. Broster (1939) advocates the high kidney incision for the removal of adrenal cortical neoplasms, whereas Crile (1923) and Young (1937) prefer the oblique lumbar approach.

In the present case an oblique lumbar incision was extended medially to the linea semilunaris and the peritoneum was opened widely. The suprarenal vein, which was greatly enlarged, and the renal vessels were ligated after turning the second part of the duodenum medially. No attempt was made to separate the kidney from the tumour, which appeared to be invading it at the upper pole.

The survival-rate in the reported cases of this type of tumour is low. This is due to the initial lack of symptoms, and the tendency for the tumour to metastasize before causing symptoms. Most of the patients die either within a short time of appearing for treatment, or soon after operation. Cahill and others (1942), however, report a patient who presented with metastases two years after operation, and died six months later. Cottler (1948) records a patient who presented with metastases two-and-a-half years after operation, and who had survived for five years following treatment with radiotherapy.

CONCLUSIONS

An adrenal cortical carcinoma without endocrine changes is reported in a woman aged 56 years.

The tumour showed extensive calcification radiologically.

Such neoplasms should be considered in the differential diagnosis of calcification in the right or left hypochondrium.

A deep-seated mass causing distortion of the duodenum and pain can present difficulty in diagnosis in the upper abdomen.

I would like to thank Professor Sir James Paterson Ross for his very kind help, advice, and permission to publish this case. I am also indebted to Mr. F. F. Rundle for his helpful criticism; Dr. Cureton for his report on the tumour and for the microphotograph; and the staff of the Photographic Department for the illustrations.

REFERENCES

ALEXANDER, R. E. (1944), *Med. Bull. Veterans' Adm., Wash.*, **20**, 341.
BROSTER, L. R. (1939), *Brit. J. Surg.*, **26**, 925.
CAHILL, G. F., MELICOW, M. M., and DARBY, H. H. (1942), *Surg. Gynec. Obstet.*, **74**, 281.
CECIL, H. L. (1933), *J. Amer. med. Ass.*, **100**, 463.
CLEVELAND, M., and KNOX, L. C. (1943), *Arch. Surg.*, **47**, 192.
COTTLER, Z. R. (1948), *J. Urol.*, **60**, 363.
CRILE, G. W. (1923), *Ann. Surg.*, **88**, 470.
HERMANN, L. (1943), *J. Urol.*, **50**, 550.
KAPLAN, J. H., and GREENE, L. F. (1948), *Surg. Clin. N. Amer.*, **28**, 1071.
KELYNACH T. N. (1897), *Med. Chron.*, **7**, 401.
KEPIER, E. J., and KEATING, F. R. (1941), *Arch. intern. Med.*, **68**, 1010.
LOEB, M. J. (1941), *J. Urol.*, **45**, 785.
LUKENS, F. D. W., FLIPPEN, H. F., and THIGPEN, F. M. (1937), *Amer. J. med. Sci.*, **193**, 812.
ROLLESTON, H. D., and MARKS, H. W. J. (1898), *Ibid.* **116**, 383.
SAMUEL, E. (1948), *Brit. J. Radiol.*, **21**, 139.
SHENTONE, N. S., and LOUGHEED, G. W. (1936), *Canad. med. Ass. J.*, **34**, 188.
WILLIS, R. A. (1948), *Pathology of Tumours*. London: Butterworth.
WU, H. H. (1940), *Chinese med. J.*, **3**, 52.
YOUNG, H. H. (1937), *Genital Abnormalities, Hermaphroditism and Related Adrenal Diseases*. London: Baillière, Tindall & Cox.

A CASE OF ACUTE INSUFFICIENCY OF SMALL INTESTINE

WITH A DISCUSSION AND REVIEW OF THE LITERATURE

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IT is well known that patients who have had massive resections of the small intestine, usually for mesenteric thrombosis or embolism, can support life adequately on a surprisingly small amount of jejunum. This paper reports the history of a man who had about one-quarter of the upper end of his small intestine short-circuited for benign fibrous strictures. As a result of this operation he came

near to dying of starvation, showing gross loss of weight, fatty diarrhœa, and hypoproteinæmia, in spite of an adequate intake of food by mouth. Following an operation in which the short-circuit was unpicked, he made a rapid and dramatic recovery.

CASE REPORT

HISTORY.—A man, aged 25, was admitted to St. Thomas's Hospital under Mr. N. R. Barrett in August, 1948.

His previous history was that in September, 1947, he had been operated upon as an emergency at another hospital for subacute obstruction. At this operation two localized fibrous strictures of the jejunum were found about 2 ft. apart, and a short-circuiting operation was performed. Following this operation the wound became septic and his feet and face became swollen. He was discharged, and in December, 1947, was admitted to another hospital where an empyema was drained.

FIG. 412.—Photographs of patient before operation. Note œdema of legs and scrotum. Ascites. Thin scraggy arms.

On admission to St. Thomas's, one year after the abdominal operation, his complaints were: (1) diarrhœa (his bowels were open 2-hourly night and day; the stools were loose, offensive, and light coloured; this diarrhœa in varying degree had been present ever since the operation); (2) colicky central abdominal pain; he had lost over 2 st. in weight over the last six months, and his legs had become very swollen for the last three months.

ON EXAMINATION (*Fig.* 412).—He was pale, thin, and looked like the inmate of a Japanese P.o.W. Camp. He had ascites, œdematous scrotum and feet, and was extremely weak. There was a sinus in the lower part of the left chest, leading to a small chronic empyema cavity. His fingers were clubbed. His temperature, pulse, and respiratory rate were normal.

Investigations were as follows:—

Hb 94 per cent.

White blood-count 5,250.

Fæces: Total fat 63 per cent, of which 38 per cent is split.

Blood-urea 22 mg. per cent.

Urine: Normal, no albumin or sugar.

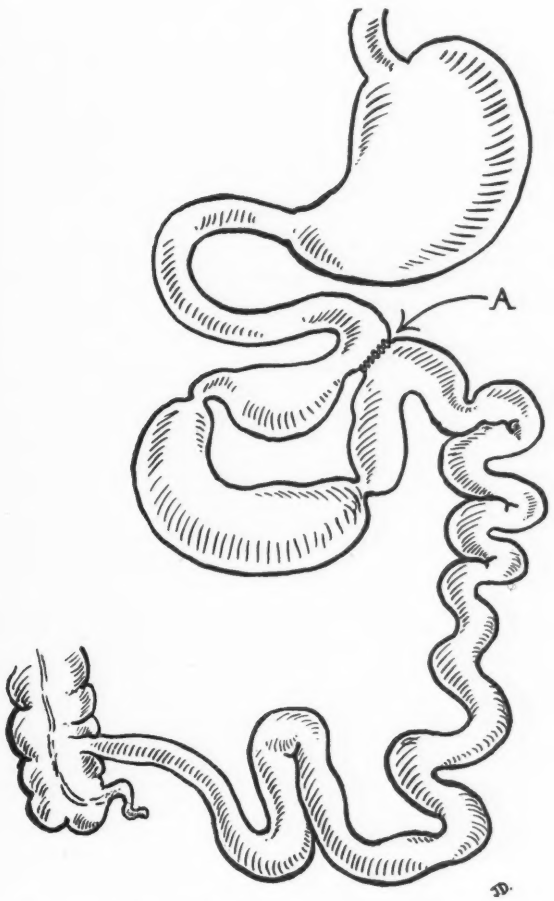

FIG. 413.—The condition at operation, showing the two jejunal strictures and the anastomosis (A).

Serum-protein:—

First estimation		*Second estimation*	
Total	3·5 g. per cent	Total	3·7 g. per cent
Albumin	2·0 g. „ „	Albumin	1·8 g. „ „
Globulin	1·5 g. „ „	Globulin	1·9 g. „ „

The empyema was cultured and from it *B. proteus* was grown. Three weeks after admission his empyema was re-operated upon and adequately drained. A biopsy of the walls taken at the time showed no evidence of tuberculosis.

A barium meal was next attempted, but was not a success as the patient was too weak to co-operate; the radiograph showed several coils of very distended small intestine in the upper abdomen. He appeared to be going downhill very rapidly at this time and laparotomy was decided upon, as it was evident that he would soon be too weak to withstand an operation.

AT OPERATION.—On Sept. 7, 1948, under general anæsthesia, the abdomen was opened. About four or five pints of clear watery liquid came out. A very large and distended closed loop of jejunum was revealed, which contained two benign strictures about 2 ft. apart. The short-circuit was within an inch of the duodeno-jejunal flexure, and appeared to involve a length estimated as approximately a quarter of the total length of the small intestine (*Fig.* 413). The whole intestine was œdematous; the stoma marked A in *Fig.* 413 was patent and the mesentery was full of small glands. The anastomosis at A was unpicked, and the two strictures by-passed by small local short-circuits (*Fig.* 414). A gland from the mesentery was taken for biopsy. This showed non-specific inflammatory change on section.

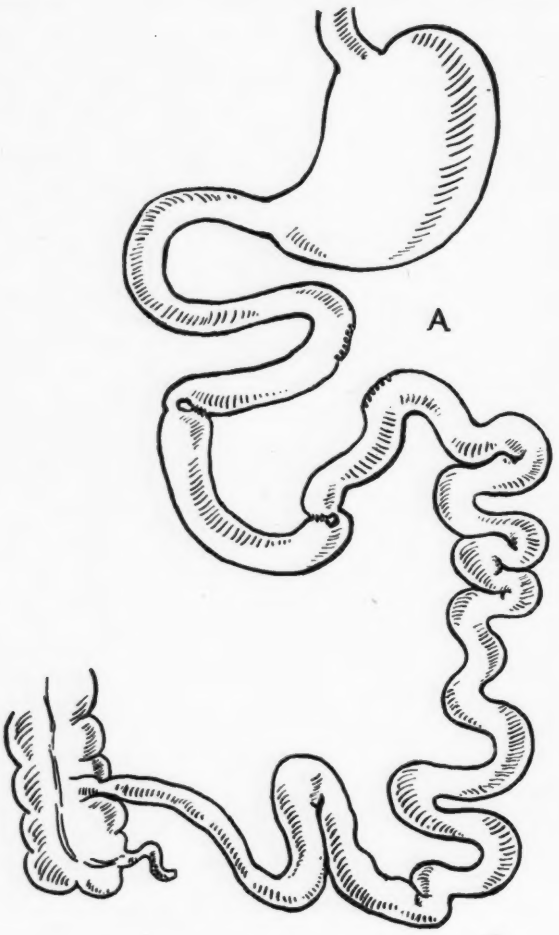

FIG. 414.—The position at the end of operation. The original anastomosis was unpicked and the two strictures by-passed by local short-circuits.

PROGRESS.—On the sixth post-operative day he burst his abdomen and this had to be resutured by strong through-and-through silkworm gut sutures.

His period of recovery from this operation presented three interesting features: First, rapid relief from all his symptoms. Pain ceased immediately. Diarrhœa decreased at once, and two weeks after the operation he was having his bowels open twice a day only. Three weeks after the operation examination of stool showed total fat 58 per cent (70 per cent split). Secondly, his œdema subsided rapidly. Three weeks after operation there was no clinical evidence of œdema and his blood-protein was:—

Total 5·4 g. per cent	Albumin	1·6 g. per cent
	Globulin	3·42 g. „ „
	Fibrinogen	0·38 g. „ „

Three weeks later the values were normal, being:—

Total 7·5 g. per cent	Albumin	3·92 g. per cent
	Globulin	3·58 g. „ „

Finally, during the month after operation, when he was rapidly putting on weight, his pulse showed a persistent rise from the fairly steady pre-operative level of between 80 and 95 per minute to between 100 and 120 per minute. This level was maintained for the whole month, and then gradually fell to normal. It caused some concern, until it was pointed out that this persistent tachycardia also occurred in people who were recovering from starvation on an adequate protein diet. It is thought to be due to the general rise of metabolic rate caused by the specific dynamic action of the increased protein metabolism. *Fig.* 415 shows the patient one month after operation.

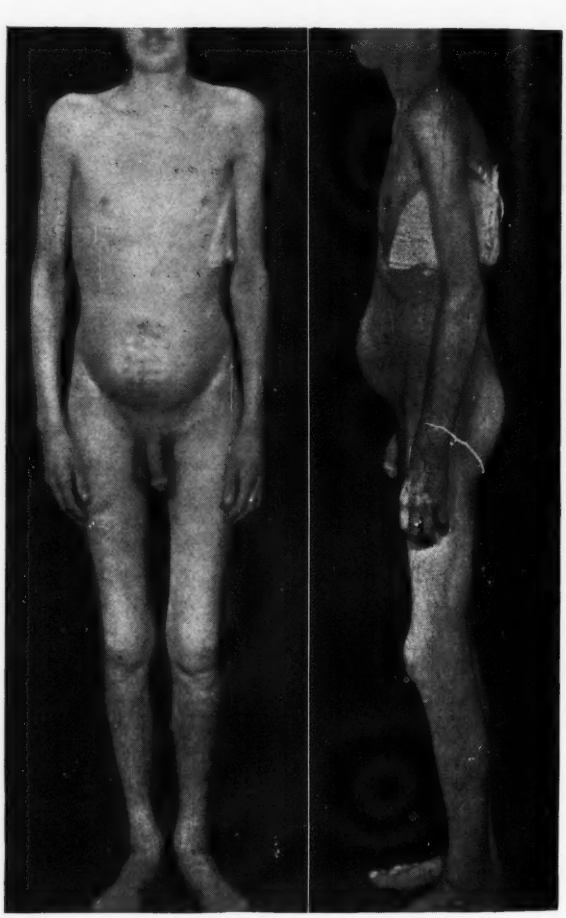

FIG. 415.—Four weeks after operation. All œdema has gone. Nutrition has improved, ribs do not show so prominently. Abdominal bulge is now due to ventral hernia, not ascites.

The patient was last seen three and a half months after his operation, when he was in every way normal and just starting his job again.

DISCUSSION

Why did this man show all the signs of severe and acute nutritional deficiency, and why was

recovery so complete and rapid after the operative procedure outlined above?

It cannot have been due to the fact that one-quarter of the small intestine was out of action, even though that quarter was jejunum—notably the area which is most adapted for absorption. There have been well over 300 cases recorded in the literature where massive resection of the small intestine has been successfully carried out. Often no more than 2–3 ft. of jejunum, anastomosed to transverse colon, has been left. H. E. Haymond (1935) reviewed 257 such cases, and it is notable that in survivors none showed nutritional deficiency or symptoms in any way comparable to those recorded here. The only residual symptom in most cases was a mild fatty diarrhœa—usually very adequately controlled by limiting dietary fat. Hypoproteinæmia has not been recorded after such operation.

The surgical error of gastro-ileostomy might be expected to present a parallel to the present case. Twenty-two cases only have been reported in the literature, and they have recently been reviewed by Brown, Colvert, and Brush (1947). Again, in no case was the nutritional deficiency anything like as severe as in our case. Loss of weight, diarrhœa and vomiting, and occasionally pain (due to secondary ulcer) are the leading symptoms of this condition. Severe hypoproteinæmia was not recorded.

The only parallels to the present case would appear to be found in certain examples of Crohn's disease of the small intestine. Often the rapid loss of weight, diarrhœa, and hypoproteinæmia shown by these patients is out of all proportion to the abnormality present. The explanation is thought to be that the area of Crohn's disease acts as an 'irritable focus', setting up reflex hyperperistalsis in the whole of the gastro-intestinal tract. In other words, food is not allowed to remain in the small intestine long enough to be digested or absorbed.

Now in considering the present case, the essential lesion found at operation was a large closed loop of jejunum, containing two strictures; this was *distended* and it was at the upper end of the small intestine. Distension is the physiological stimulus to the bowel. It is known, for instance, that if a balloon is placed in the jejunum and distended, it excites hyperperistalsis so that it is rapidly passed along the intestinal tract. The use of the Miller-Abbott tube depends on this fact. Thus, this large distended loop of jejunum must have acted as a powerful 'irritable focus' causing gross reflex gastro-intestinal hurry. The fact that the loop itself consisted of jejunum, where a great deal of normal absorption takes place, must have enhanced the nutritional difficulty. The dramatic recovery following simple elimination of the large distended closed loop (but leaving the strictures locally short-circuited only) supports the explanation outlined above.

The case might also act as a warning against a surgical procedure which leaves a large loop of small intestine in situ which is liable to become distended.

I am indebted to Mr. N. R. Barrett for his help, and for permission to report this case.

BIBLIOGRAPHY

BROWN, C. H., COLVERT, J. R., and BRUSH, B. E. (1947), *Gastroenterology*, **8**, 70.
COGSWELL, H. D. (1948), *Ann. Surg.*, **127**, 377.
FLINT, J. M. (1912), *Bull. Johns Hopk. Hosp.*, **23**, 127.
HAYMOND, H. E. (1935), *Surg., Gynec. Obstet.*, **61**, 693.
HOLMAN, C. C. (1944), *Lancet*, **2**, 597.
STANDARD, S. (1946), *Amer. J. digest. Dis.*, **13**, 320.
WILDEGANS, H. (1925), *J. Amer. med. Ass.*, **85**, 1, 436.

THE TREATMENT OF SUPPURATIVE ARTHRITIS OF THE INTERPHALANGEAL AND METACARPOPHALANGEAL JOINTS*

BY A. G. RIDDELL, M.B.E.
LATE JOHN MARSHALL FELLOW IN SURGICAL PATHOLOGY

SUPPURATIVE arthritis of the joints of the fingers is a relatively uncommon condition. A study of the notes of the last two thousand patients attending the Hand Clinic of University College Hospital has revealed only 5 cases. Notwithstanding its infrequent occurrence it is an important condition because it is likely to cause a prolonged period of disability, followed, when healing occurs, by a stiff finger. In order that the disability period may be reduced, immediate amputation of any digit except the thumb has been advocated. (Bunnell, 1948.)

Experience in treating infections of the phalanges has shown that, if the causal organism is sensitive to penicillin and the sequestrum small, the lesion may be sterilized by giving systemic penicillin over a long period. In suppurative arthritis there is little dead bone, but there is necrotic cartilage which acts as a foreign body and causes a prolonged purulent discharge from a sinus. The method of treatment adopted has been to operate on cases of suppurative arthritis as soon as possible after the diagnosis has been made. The operation consists of a joint toilet followed by primary closure of the wound. The latter is made possible by the use of systemic penicillin.

The case notes of the five cases treated by this method are presented below.

* From the Surgical Unit, University College Hospital Medical School.

CASE REPORTS

Case 1.—C. Y. was admitted to University College Hospital on May 1, 1946, fourteen weeks after a crush injury with laceration of the skin over the distal interphalangeal joint of the right index finger. Fourteen days before admission he had attended the Hand Clinic, when the lesion had healed but the two distal segments

of his finger were painful, swollen, and red. An exploratory incision was made but no pus was found. A radiograph taken at this time (April 16) showed narrowing of the joint space but no destruction of bone. The wound failed to heal and discharged serous fluid, a culture of which grew *Staph. pyogenes aureus* and hæmolytic streptococci. A further radiograph on April 29 showed a small sequestrum.

Examination on the day of admission showed a diffuse swelling of the distal two segments of the index finger with two small sinuses over the distal interphalangeal joint, from which clear fluid was exuding. Movement of the distal interphalangeal joint was painful and there was lateral instability.

TREATMENT.—Intramuscular penicillin 30,000 units was given three-hourly from May 1 to May 13.

Operation was performed on May 1 under brachial plexus block with a bloodless field. The sinuses were excised together with an elliptical area of skin over the dorsal surface of the joint. The granulation tissue overlying and in the joint was gently curetted. No loose cartilage or bone was found. The skin was closed with interrupted silk sutures and a plaster splint was applied.

On May 6 the wound was healed and the sutures were removed. The patient was discharged from hospital on May 14 and he returned to work on May 31. The lesion was then painless, but he had no active movement in the affected joint.

Case 2.—S. V. first attended the Hand Clinic on Dec. 9, 1946. Eighteen days previously he had caught his left index on a wooden splinter. The wound was over the dorsal surface of the proximal interphalangeal joint. Two days after the injury the wound had become painful and a week before attending the clinic it had begun to discharge pus.

Examination showed a diffuse swelling of the middle segment and distal half of the proximal segment of the finger, which were swollen to twice their normal size. A sinus was present over the dorsal surface of the proximal interphalangeal joint, from which pus was exuding. Crepitus was present on moving the joint.

A radiograph of the affected joint on Dec. 9 showed loss of the joint space and marked rarefaction of the adjacent phalanges.

TREATMENT.—The patient was admitted on Dec. 12 and intramuscular penicillin, 30,000 units three-hourly, was given from the day of admission until Dec. 30.

A culture taken on Dec. 16 was sterile, but a smear showed a few Gram-positive cocci.

On Dec. 17 operation was performed. The sinus was enlarged and the joint opened. An abscess cavity was found on the volar surface of the joint. This abscess cavity and the joint were gently curetted. The lateral half of the extensor tendon was found to be intact. The skin was closed with interrupted silk sutures.

The sutures were removed on Dec. 25, when the wound was soundly healed. The patient was discharged from hospital on Dec. 30.

This patient's out-patient notes have been lost, but he was seen again in January, 1949. There is bony ankylosis of the proximal interphalangeal joint in 20° of flexion. The scar is not tender. He has a full range of movement at the metacarpophalangeal and distal interphalangeal joints.

Case 3.—R. B. was admitted with the history that four weeks previously he had cut the dorsal surface of the ring finger over the distal interphalangeal joint with a razor-blade. The wound failed to heal and after two weeks became painful and began to discharge pus. He was treated elsewhere with hot baths and a sulphonamide powder for two weeks. During this time the finger became more painful and he was then referred to the Hand Clinic and was admitted as an emergency on July 2, 1947.

On examination there was diffuse swelling of the distal two segments of the ring finger, most marked over the extensor surface of the terminal segment. A small sinus was present over the dorsal surface of the distal interphalangeal joint down which a probe could be passed into the joint cavity. Movement of the joint was not painful.

An X-ray examination showed rarefaction and necrosis of the joint surface. A culture taken the day of admission grew *Staph. pyogenes aureus* which was penicillin-sensitive.

TREATMENT.—Intramuscular injections of penicillin, 100,000 units three-hourly, was commenced on July 2 and continued until July 10. Sulphamezathine, 1 g., was given four-hourly for three days from July 3, while the report on the culture was awaited.

On July 3 operation was performed under brachial plexus block with a bloodless field. The sinus was excised and the skin incision prolonged to give adequate exposure of the joint. The extensor tendon was found to be divided. Periarticular granulation tissue and necrotic cartilage were gently curetted. The divided extensor tendon was sutured with interrupted sutures of ophthalmic silk and the skin closed with interrupted silk.

The hand was immobilized with a plaster splint. The sutures were removed on July 10. The wound had healed by first intention. The patient then had a range of 10° painless movement in the joint. It was intended to give the patient physiotherapy as an out-patient, but he insisted on returning to work.

The patient was not seen again until March, 1948. He considered that the finger was normal in every way. Examination showed slight periarticular thickening. Extension was full at the distal interphalangeal joint and 30° of active flexion was present. One silk suture from the tendon was ulcerating the skin and was removed.

Case 4.—W. C. was admitted as an emergency on Sept. 16, 1948. A week previously while washing a broken glass he cut his right thumb over the dorsal surface of the interphalangeal joint. Since the accident the thumb had been very swollen and painful and the wound had discharged clear fluid.

Examination on the day of admission showed gross swelling of the whole thumb. There was a gaping wound 2 cm. long. Movement of the interphalangeal joint was extremely painful.

TREATMENT.—Intramuscular injections of penicillin 150,000 units three-hourly were commenced on Sept. 16, and continued until Oct. 1. Sulphadiazine was given orally from Sept. 16 to Sept. 25, the total dose being 61 g.

On the day of admission the wound was explored under a ring block and it was found that a probe could be passed through a small hole in the extensor tendon into the joint, which contained pus. A small diamond-shaped piece of tendon was excised to allow drainage of the joint and through this penicillin was injected into the joint. The wound was not closed and the whole hand was immobilized on a plaster splint.

The wound was dressed daily and on alternate days a ring block was given and penicillin injected into the joint.

By Sept. 25 the soft-tissue swelling had subsided sufficiently for an attempt at suture to be made. Under ring block a dorsilateral incision was made on the ulnar side of the joint. The extensor tendon was reflected towards the radial side of the finger to allow exploration of the joint cavity and several loose fragments of cartilage were removed. The sinus was excised and both wounds were closed with interrupted sutures of stainless steel wire.

On Oct. 2 the sutures were removed and the wounds were well healed. The patient was discharged from hospital and attended the Department of Physical Medicine for active exercises. On Oct. 18 the patient returned to work. He then had 20° of active flexion at the interphalangeal joint, which was painless.

Case 5.—H. H. attended the Hand Clinic on Aug. 30, 1948, with the history that a fortnight before he had run a piece of wire into the dorsum of his left hand adjacent to the metacarpophalangeal joint of the ring finger. The dorsum of his hand became swollen and painful.

Examination showed a generalized œdema of the dorsum of the hand. A scar 1 cm. long with a scab on it was present between the heads of the third and fourth metacarpals. A tender fluctuant area 2 cm. in diameter was present over the head of the fourth metacarpal. Movement of the metacarpophalangeal joint was not painful.

An incision was made above this tender area, but no pus was found. The patient was given penicillin 500,000 units twice daily for the next forty-eight hours.

A radiograph taken the next day showed minor changes in the fourth metacarpophalangeal joint which were thought to be due to osteo-arthritis.

The wound healed by Sept. 6 and the patient was transferred to the Department of Physical Medicine with a diagnosis of osteo-arthritis of the metacarpophalangeal joint. He was treated with active exercises and short-wave diathermy until Sept. 21. His condition was then very much worse and the whole hand was swollen and painful. A second radiograph showed osteoporosis of the whole hand, with narrowing of the joint space of the fourth metacarpophalangeal joint. There was necrosis of the head of the metacarpal and subperiosteal new-bone formation along its shaft.

The diagnosis of suppurative arthritis was then made and he was admitted on Sept. 26.

TREATMENT.—Systemic penicillin 100,000 units three-hourly was given from Sept. 27 to Oct. 4.

On Sept. 29 operation was performed under a wrist block with a bloodless field. A dorsilateral incision 5 cm. long was made on the ulnar side of the joint and the extensor tendon was mobilized towards the radial side to allow exploration of the joint. The synovial membrane was œdematous and the cartilage on the head of the metacarpal was necrotic. The necrotic cartilage was excised and the joint capsule closed with interrupted sutures of No. 43 stainless steel wire. The skin was closed with interrupted stainless steel wire. The whole hand was immobilized with a plaster splint.

The sutures were removed on Oct. 8 and the wound was soundly healed. The patient was sent for further physiotherapy.

The recovery in this patient has been slow; when seen on Jan 12, 1949, the functional recovery was very poor. He had regained 70° of painless flexion at the metacarpophalangeal joint but he had poor co-ordination of the whole hand.

DISCUSSION

Each of these cases of suppurative arthritis has resulted from a wound that penetrated the joint capsule. Other causes of suppurative arthritis are only met with occasionally. One case of suppurative arthritis resulting from a spreading infection with suppurative tenosynovitis has occurred at this clinic. This case has not been included in the series as its treatment should be discussed with that of suppurative tenosynovitis. A few cases of joint effusion accompanying subcutaneous abscesses on the volar surface of the finger have been observed, but these have always resolved without suppuration after the administration of penicillin. No case of metastatic arthritis has been seen.

Bacteriological investigation was not carried out on all our cases, but in three the causal organism was a Gram-positive coccus. It is probable that the other two cases were staphylococcal in origin as other organisms are rarely met with in hand infections. An exception to this generalization is the suppurative arthritis of the metacarpophalangeal joints following human or animal bites; in these cases a varied flora has been reported.

The diagnosis of suppurative arthritis is often difficult to establish and the picture may be masked to some extent by chemotherapy. The most important physical signs are a wound in the region of a joint, particularly on the dorsal surface, swelling of the soft tissues in the region of the joint, and painful limitation of movement. However, movement of the affected joint is not always painful (*Cases* 3 and 4). Wounds over the dorsal surface of the finger-joints should always be carefully explored to exclude damage to the joint capsule, and while this is being done the finger should be placed in the position it was in at the time of the original injury, so that a lesion of the joint capsule may be more easily demonstrated. If doubt exists as to the diagnosis of suppurative arthritis the lesion should be explored under local anæsthesia (*Case* 4).

Case 5 demonstrates the difficulty that may be experienced in arriving at the correct diagnosis. The correct diagnosis was considered when the patient was first seen and the first operation was undertaken with the expectation that either there was suppurative arthritis present or that an abscess would be found in close relation to the joint. The failure to find such a lesion was misleading and the rapidity with which the wound healed confused the clinical picture still further. We have twice seen cases of osteo-arthritis of the metacarpophalangeal joint of the thumb aggravated by trauma which were at first suspected of being cases of suppurative arthritis; in both instances the lesion rapidly improved after a few days' immobilization on a plaster splint.

It is adequate chemotherapy that has made this operation possible. In the early cases the dose of penicillin was 30,000 units three-hourly, but our experience in the treatment of infections of the phalanges suggests that the dose should be greater than this and the more recent cases have been given a higher dosage, usually 100,000 units three-hourly, and in one case 150,000 units three-hourly. *Cases* 3 and 4 were also given a course of sulphonamide therapy. This was unnecessary in *Case* 3 and probably unnecessary in *Case* 4, but in the latter it was continued because the causal organism was not isolated.

The operation should be performed as soon as possible after the diagnosis has been made. Operation had to be delayed in *Case* 4 because the swelling of the soft tissues would have made it impossible to excise the wound and suture it.

The operation must be performed in a bloodless field. Any form of anæsthesia may be used, but either a ring block or a brachial plexus block with a sphygmomanometer used as a pneumatic tourniquet has proved satisfactory.

The operative approach used depends on the degree of injury to the extensor tendon. If this has been more than half divided a dorsal approach including the sinus present may be used. If the extensor tendon is undamaged or has only a small puncture wound in it, a dorsilateral incision with mobilization of the tendon should be used to obtain access to the joint cavity (*Cases* 4 and 5). Great care must be exercised to avoid damage to the lateral ligaments of the joint.

Exploration of the joint itself is awkward because the joint surfaces can be separated very little from one another and the inspection of the volar aspect of the joint is difficult. This is particularly so in the case of the metacarpophalangeal joint when there is marked convexity of the head of the metacarpal. In suppurative arthritis the synovial membrane may be ballooned out on the volar aspect of the joint to contain the main abscess cavity (*Case* 2).

The diseased cartilage and bone must be carefully removed. Rough handling of the joint surfaces will result in ankylosis of the joint.

In those cases in which the joint is not completely disorganized, the extensor tendon, if damaged, should be repaired. The method recommended by Bunnell (1948) using removable figure-of-eight stainless-steel wire sutures through both the tendon and the skin has not been found to be satisfactory. With this method the lines of suture in both tendon and skin inevitably lie in contact, and they adhere. Suture of the tendon should be performed separately with buried sutures. The finger can then be immobilized in such a position that the two suture lines are not superimposed. In *Case* 3 suture of the tendon was performed with ophthalmic silk. Silk, however, does produce a slight tissue reaction, and in this case one of the sutures was later extruded. The tendon should be sutured with fine interrupted sutures of stainless steel wire which will not evoke a tissue reaction.

After completion of the operation the whole hand should be placed on a plaster slab to immobilize all the digits. It is not sufficient to apply a plaster splint to one finger alone.

When the wound is firmly healed active exercises should be commenced and practised under the guidance of a physiotherapist.

In all five cases joint toilet and suture of the wound has been followed by first-intention healing. In three cases (*Cases* 3, 4, and 5) a useful range of active movement has been regained in the damaged joint. The results are summarized in the accompanying table.

DETAILS OF 5 CASES OF SUPPURATIVE ARTHRITIS

CASE NUMBER	JOINT AFFECTED	CAUSE OF DISABILITY	TIME FROM INJURY TO ATTENDANCE AT HAND CLINIC	TIME FROM FIRST ATTENDANCE TO WOUND HEALING	TIME FROM FIRST ATTENDANCE TO RETURN TO WORK	RESULT
1	Distal interphalangeal joint of index	Crush injury	84 days	20 days	44 days	Painless fixed joint
2	Proximal interphalangeal joint of index	Wooden splinter	18 days	17 days	Not known, approximately 21 days	Painless fixed joint
3	Distal interphalangeal joint of ring finger	Cut with razor blade	28 days	9 days	10 days	30° active flexion present
4	Interphalangeal joint of thumb	Cut with glass	7 days	17 days	33 days	20° active flexion present
5	Metacarpophalangeal joint of ring finger	Puncture wound with a piece of wire	14 days	40 days	More than 136 days*	70° active flexion present

* Waiting to attend Compensation Tribunal.

That the wound healed rapidly following treatment is very satisfactory, but the total disability period in all cases has been prolonged. Examination of the table will show that this is due to :—

1. Delay in commencing treatment.
2. Delay in making the diagnosis of suppurative arthritis.
3. Delay in return to work because physiotherapy may be required for a considerable period.

However, all these cases if left untreated would have had a painful finger with a discharging sinus for many months, which would have inevitably led to an ankylosed joint if not to amputation of the finger. *Case* 5 could have returned to light work in approximately eight weeks from the time of his first attendance and the others all returned within 44 days.

These results can be improved upon, but they demonstrate that the prognosis in suppurative arthritis of the joints of the fingers is not hopeless and the method of treatment may give a considerable degree of functional recovery.

SUMMARY

1. Five cases of suppurative arthritis of the joints of the fingers are reported.
2. Rapid healing was achieved in all cases by joint toilet and suture of the skin with concurrent pencillin therapy.
3. In three cases a useful recovery of movement in the affected joint was obtained.

REFERENCE

BUNNELL, S. (1948), *Surgery of the Hand.* Philadelphia.

INFERIOR MESENTERIC VASCULAR OCCLUSION

By J. Y. W. RUSSELL, Preston

Occlusion of the superior mesenteric artery in its trunk or in its distribution is a relatively common condition and there are numerous recorded cases. In the literature reference is frequently made to occlusion of the inferior mesenteric artery, but case reports are few and it would appear that it is quite a rare condition.

Desplas (1927) recorded one case in which the large gut was involved from transverse colon to sigmoid. In the American literature cases have been recorded by MacCormack (1932), Harkins (1936), who found that only one out of nine cases of mesenteric vascular occlusion occurred in the inferior mesenteric distribution, and Laufman and Scheinberg (1942), in whose series of 44 cases only 2 were inferior mesenteric. Charles Mayo, Miller, and Stalker (1940), in discussing mesenteric vascular occlusion, speak of the mesenteric artery and do not mention the inferior vessel. This omission is also common in many of the standard text-books.

The following case is therefore of considerable interest.

CASE REPORT

History.—The patient, a widow of 73, was admitted to the Medical Wards of the Preston Royal Infirmary on April 18, 1948. She had been known to have diabetes for some seventeen years, but had consistently refused insulin therapy. She was now in a state bordering on diabetic coma and had an early gangrene of the right foot. Stabilization of the diabetic condition with insulin therapy combined with chemotherapy did not fully arrest the condition and a below-knee amputation was carried out on Aug. 17, (R. S. G.). Notwithstanding her poor general state, she tolerated this intervention very well and, apart from a slight intermittent pyrexia, her convalescence proceeded uneventfully until Aug. 26. She took her lunch quite well, but shortly afterwards complained of severe lower abdominal pain of sudden onset and vomited twice. The vomitus consisted only of gastric contents. This was followed by repeated retching. The pain continued to be severe, with occasional exacerbations. There was no associated backache or shoulder pain. There was free perspiration. The bowels were incontinent, with almost continual passage of loose brownish motions with no admixture of blood or slime. The bowel action had previously been quite regular, but the patient stated that for the last two months she had passed a little fæcal matter through the vagina. She had borne seven children without any great difficulty and had no previous pelvic operations or treatment. There had been no urinary symptoms.

On Examination.—The patient was pale and shocked and evidently in great pain. The pulse rate was 120 and the temperature 99·8°. The tongue was moist and lightly coated. The abdomen was a little distended and across its lower part there was considerable tenderness and rigidity, which was a little more marked on the left side. The epigastrium was quite soft and not tender. Bowel sounds were absent. There was no loss of liver dullness. Vaginal examination revealed some tenderness in the pouch of Douglas. Fæces were present in the vagina, and on the posterior wall there was a small fistulous opening into the rectum. The edges of this opening were smooth, with no thickening. Rectal examination revealed a great amount of fluid brown fæcal matter to be present and confirmed the presence of the fistula. No blood or slime and no other abnormality of the rectal wall was detected.

Diagnosis.—The acute onset and marked peritonism suggested a perforated viscus. Because of the poor state of the patient's vascular tree, intra-abdominal thrombosis was considered to be a possibility.

Immediate operation was advised, but the patient declined and there was a delay of some hours while a relative was brought to hospital and carried out the necessary persuasion. Intravenous saline was given during this waiting period. It was considered that the administration of an enema would be unwise.

At Operation.—This was commenced six hours after the onset of the symptoms. A lower right paramedian incision was made. There was a considerable amount of yellowish seropurulent fluid in the peritoneum. The small intestine, the cæcum, ascending colon, and the proximal two-thirds of the transverse colon were quite normal, but the large gut distal to this, as far down as the middle of the rectum, was flaccid and of a slaty-grey colour. There was some fluid fæcal content. The transition from normal healthy pink to gangrenous bowel was quite abrupt, especially at the junction of the middle and left thirds of the transverse colon. Resection of the non-viable bowel was carried out after mobilization in the usual way. The transverse colon was divided about its middle through healthy bleeding tissue. The ' mesentery ' of the left half of the transverse colon, the descending colon, and the pelvic colon was divided some little distance from the gut. There was found to be no need to apply clamps on the bowel side of the line of section of the mesentery as there was no ' back bleeding '. The rectum was divided between two right-angled hysterectomy clamps just above the pelvic floor and the rectal stump closed with a running suture over the clamp, which was then withdrawn and the line of closure oversewn. At this stage there was some fæcal contamination, as the rectum was quite tense with the fæcal matter which it had been considered inadvisable to evacuate prior to operation. Satisfactory closure was, however, obtained. The end of the transverse colon was clamped after being brought out through the left rectus muscle as a terminal colostomy, and the abdomen was closed with a tube drain down to the pelvic floor. A tube was inserted into the rectum and left as a drain.

Progress.—Intravenous saline and plasma and gastric suction were continued after operation and the patient's condition gradually improved. Penicillin was given in doses of 100,000 units three-hourly and soluble sulphathiazole in full doses in the saline infusion. On the day following operation the clotting time was found to be 3½ minutes and heparin and dicoumarin were administered for four days with repeated checks of the clotting time and prothrombin index.

The clamp was removed and a Paul's tube inserted into the end of the colon after 60 hours. This was rapidly followed by the passage of flatus and fæces and the colostomy continued to act well. There was some necrosis of the end of the gut, however, and the Paul's tube came out two days later. On Sept. 3, eight days after operation, the patient's condition began to regress and she became weaker. A slight icteric tinge appeared and gradually deepened. The bowel end had now retracted a little and a purulent discharge appeared alongside. Culture of this revealed staphylococci and *B. proteus*. A thick catheter was introduced into the gut for drainage purposes.

Liver function tests were carried out with the following results: Van den Bergh reaction 14 units; Thymol turbidity 3·4 units; Thymol flocculation plus; Takata Ara 0 to plus; Alkaline phosphatase 27 units; Colloidal gold 4; Alcohol turbidity 23 units. This suggested a toxic hepatitis. The jaundice deepened and the patient weakened and died on Sept. 7.

POST-MORTEM EXAMINATION (A. A. M.).—

External Examination.—The body was that of an elderly female in a poor state of nutrition. The right leg had been amputated below the knee. There was a colostomy wound in the left lower abdomen. The skin and mucous membranes of the body were intensely jaundiced.

Internal Examination.—

Lungs and pleuræ: Both lungs were œdematous and showed acute passive congestion.

Heart and vessels: The right side of the heart was dilated and the left contracted. There was evidence of advanced coronary atheroma, which was also present in the aorta. The proximal part of the inferior mesenteric artery was markedly atheromatous and showed almost total occlusion of its lumen within 1 cm. from its origin.

Abdomen: The peritoneal cavity contained a small amount of creamy foul-smelling pus. There was evidence of acute fibrinous peritonitis. The descending colon, pelvic colon, and rectum had been removed at operation. The mesentery of the pelvic colon and the colostomy opening in the transverse colon were gangrenous. The remainder of the colon was dark and almost black in colour. The small intestine was normal except for the presence of peritonitis.

Liver: This was of normal size and consistency. On section numerous yellowish patches were scattered throughout its substance, probably the result of toxic hepatitis.

Kidneys: These showed the macroscopical appearances of chronic nephritis.

HISTOLOGY.—

Liver: The parenchyma showed marked cloudy swelling and granular degeneration with fatty infiltration. Many of the nuclei were enlarged, pale, and vacuolated. The portal tracts showed perivascular round-cell infiltration; in some areas there was increase of connective tissue.

Kidney: The histological picture was that of senile nephrosclerosis or arteriosclerotic kidney.

DISCUSSION

Many authors have fully described the clinical features of mesenteric vascular occlusion. It is agreed that for some time after the onset of the condition the symptoms are predominant, with acute abdominal pain as the most striking feature. Dunphy (1936), discussing this particular symptom of pain, compares it to the pain of angina pectoris. He thinks it is attributable to the anoxæmia of the intestinal musculature and is a true visceral pain, conducted by the sympathetic nervous system. Abdominal tenderness and guarding are not usually marked until much later—until, as Dunphy states, peritonitis develops. Harkins, who made a very large review of the literature, has stated that the flaccidity of the abdomen is in contrast to the pain. In this case, rigidity was present when the patient was examined soon after the onset of the pain. This suggests that the disease process might have been under way for longer than is indicated by the duration of symptoms; that a slow quiet thrombosis might have been proceeding for some hours prior to the time of the apparent acute onset, which would coincide with the development of peritonism. Nevertheless some symptoms, however mild, would have been expected, and there were none. The more likely explanation is that in inferior mesenteric thrombosis where the large gut alone is involved, peritonism is of much more rapid onset than in the superior type where small gut is usually infarcted. The wall of this is thicker and its contents less infective.

The acute onset, the elevation of pulse and later of temperature, and the absence of audible peristalsis, tended to suggest a diagnosis of perforation of a viscus in the lower abdomen, although there had been no previous alimentary symptoms to suggest any bowel disorder. Because of this an enema was not given. Cokkinis (1926) speaks of the value of a diagnostic enema in mesenteric thrombosis and states that hæmatemesis or melæna occurs in about 50 per cent of cases. Usually some of the blood in the infarcted segment is propelled into the large gut before ileus sets in and can be washed out by a high enema. Mayo states that occult blood is uniformly present in the stools in the venous type of thrombosis. Melæna was present in 13 out of 20 of Elliott's cases (quoted by Harkins, 1936). An enema in this case would have made operative manipulation easier and safer, but it is unlikely that it would have aided the diagnosis, as no blood was observed in the copious loose motions which were passed.

In the vast majority of cases of mesenteric vascular occlusion the infarcts are hæmorrhagic. There is a great outpouring of serosanguineous fluid into the peritoneum and into the gut. The bowel wall is purplish in colour, thickened, and œdematous, so that it has been likened to a tube of rubber (Desplas, 1927), and has its lumen greatly reduced. There is usually also great œdematous thickening of the mesentery. It is generally agreed that the congestion is due to inflow of blood from the periphery, and this may be so marked that the vitality of the gut may be retained. This was a case of an anæmic infarct. The bowel was flaccid and greyish and there was no blood-stained exudate. The anæmic type of infarct is much less common than the hæmorrhagic. Cokkinis (1926) states that it is excessively rare. Among 44 cases, Laufman and Scheinberg (1942) had no instances of anæmic infarction. Harkins quotes Sprengel as having reported 1 anæmic infarct in a series of 4 cases. Dunphy (1936) reported 1 case of embolic occlusion in a patient with a rheumatic carditis, which would appear to have been anæmic, as the bowel was contracted and greyish, but not yet gangrenous. There appears to be a consensus of opinion that obstruction of the inferior mesenteric artery is less serious than obstruction of the superior in addition to being less common. Boyd (1947) states that this is so as it is not a terminal artery, but adds that nevertheless obstruction is frequently followed by a hæmorrhagic infarct, although gangrene is not likely to develop. Harkins quotes Mall and Welch as stating that the inferior mesenteric artery can sometimes be obliterated without serious effects, and Illingworth and Dick (1943) state that in infarction the line of demarcation is rarely abrupt. In this case there would appear to have been a minimal

anastomosis between the upper branch of the left colic artery and the left branch of the middle colic artery. At operation the line of demarcation between healthy and ischæmic gut at the junction of the middle and left thirds of the transverse colon was quite abrupt. At the other end, the line between the distributions of the superior and middle rectal vessels was also clearly demonstrated. This lack of anastomosis and subsequent inflow from the periphery accounts for the anæmic nature of the infarction.

Aetiology.—Illingworth and Dick state that because of its small diameter and the nature of its origin, the inferior mesenteric artery is rarely entered by an embolus, but the superior mesenteric is larger and at its origin runs almost parallel to the aorta and is consequently more often occluded. They state that primary thrombosis in the mesenteric arteries is very rare. It may occur as the result of extension of atheroma of the aorta, from thrombosis arising in an aneurysm of the superior mesenteric artery, and rarely in association with thrombo-angiitis obliterans. Venous thrombosis is often associated with acute appendicitis or acute inflammation of the female pelvic organs, but may come from many other sources.

There can be little doubt that in this case the thrombosis was due to the very marked atheroma which was present in the trunk of the main vessel, the mouth of which could barely be entered by a fine probe from the aorta. The very poor state of the vascular tree, aggravated by the untreated diabetes, had already precipitated gangrene of the right foot and was now in process of producing a massive gangrene of the bowel.

Treatment.—There is general agreement that if possible this should consist of resection with anastomosis. The lines of resection should be well clear of the damaged segment, as devitalization of the gut may spread after resection. Cokkinis recommends division 1 ft. proximal and distal to the lines of demarcation, and states that up to 15 ft. of small gut can be resected successfully. An alternative is exteriorization, this being applicable particularly in cases of infarction of the large gut or in rare cases where the patient's condition is too critical to allow of any other procedure. With the advent of the anticoagulants, a third line of treatment is now available. Hendry (1948) has reported 1 case in which conservative treatment was successfully carried out after exposure of an infarcted loop of small intestine and replacement in the abdomen without resection, followed by anticoagulation therapy. He also refers to Ravdin's cases and one of Rudberg's, in which similar treatment was successful. It will, of course, be necessary in every case first to establish the diagnosis by exposure of the damaged segment, in addition to assessing its viability.

In this case the treatment carried out was the only one possible in the circumstances. Exteriorization was impossible and there was no doubt about the non-viability of the segment. The technique would have been improved by a rather more proximal division of the transverse colon, in view of the later tendency of the gut at the colostomy opening to necrose, and by section of the 'mesentery' of the colon nearer to its attachment after mobilization. It is probable that the terminal hepatitis was largely due to the absorption of toxins from the sloughing mass of mesentery, as well as from the pelvic abscess. The satisfactory closure of the rectum on the pelvic floor was difficult, but the patient's poor general condition precluded a more radical resection.

Post-operatively, heparin, with or without dicoumarin, should be administered with the object of preventing any extension of the thrombosis. Murray (1940) recommends that heparin therapy be begun even during operation and continued afterwards. Laufman considers that heparin should be given only if resection is carried out, as otherwise it causes added blood and fluid loss into the bowel and the peritoneum. It is doubtful, however, if this is so, and it would appear that the extension of the condition is of much greater importance.

SUMMARY

1. A case is described of inferior mesenteric thrombosis with the production of an anæmic infarct. Death due to toxic hepatitis occurred twelve days after resection of the left half of the colon.

2. The rarity of this condition is stressed.

3. Aetiology and treatment are briefly discussed.

I should like to express my thanks to Mr. R. Cunliffe Shaw for permission to publish this case, and to Dr. A. A. Miller who performed the post-mortem examination, Mr. R. S. Garden, and Mr. W. H. Graham for helpful criticism.

REFERENCES

BOYD, W. (1947), *Surgical Pathology*, 6th ed., 91. London: Saunders.

BEATTIE, J. M., and DICKSON, W. E. (1943), *Text Book of Pathology*, 4th ed. London: Heinemann.

COKKINIS, A. J. (1926), *Mesenteric Vascular Occlusion*. London: Baillière, Tindall & Cox.

DESPLAS, B. (1927), *Arch. Mal. Appar. dig.*, **17**, 760.

DUNPHY, J. E. (1936), *Amer. J. med. Sci.*, **192**, 108.

HARKINS, H. N. (1936), *Arch. Path.*, **22**, 637.

HENDRY, W. G. (1948), *Brit. med. J.*, **1**, 144.

ILLINGWORTH, C. F. W., and DICK, B. M. (1943), *Surgical Pathology*. London: Churchill.

LAUFMAN, H., and SCHEINBERG, S. (1942), *Amer. J. Surg.*, **58**, 84.

MACCORMACK, R. L. (1932), *Wis. med. J.*, **31**, 702.

MAYO, C. W., MILLER, J. M., and STALKER, L. K. (1940), *Coll. Pap. Mayo Clin.*, **32**, 328.

MURRAY, G. (1940), *Arch. Surg.*, **40**, 307.

A CASE OF SO-CALLED LATERAL ABERRANT THYROID

THE IMPORTANCE OF HEMITHYROIDECTOMY

By R. S. MURLEY, St. Bartholomew's Hospital, London

No apology is made for the title of this report of a case of metastasis from an occult carcinoma of the thyroid. The features of the case are important in that they convincingly demonstrate the grave danger of assuming a developmental origin for any laterally situated tumour of thyroid tissue in the neck.

The subject of the so-called lateral aberrant thyroid has attracted interest for some years and several hypotheses have been put forward to explain the occurrence of these tumours. The idea that such tumours may originate from the lateral thyroid anlagen or the ultimobranchial bodies has the strong support of no less an authority than Lahey (Lahey and Ficarra, 1946), yet this view is certainly wrong. In this respect it is perhaps significant that Crile (1939), who formerly supported Lahey's view, has recently reported 21 consecutive cases where careful examination of the thyroid gland revealed a primary tumour (Crile, 1947). The remarkably slow growth of both the primary tumour and its cervical metastases have caused much confusion in the past, nor is this surprising when one considers the extraordinarily long history in most cases.

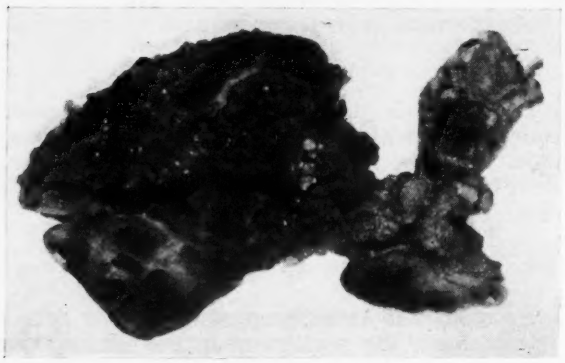

Fig. 416.—Cut surface of main metastasis and two smaller glands. Papilliferous growth is present in the large gland to the left and in one of the smaller glands. The lower gland on the right is not involved. (× 1½.)

The present case is reported because it illustrates so well the very great importance of exploring the thyroid and performing a hemithyroidectomy in every case of this kind.

CASE REPORT

History.—Mr. L. S., aged 39, an insurance clerk, was admitted to St. Bartholomew's Hospital on Jan. 21, 1948, complaining of a lump in the right side of his neck. The patient stated that this had appeared five years before; that it had enlarged quite suddenly at first and then remained almost stationary in size. There were no other symptoms and the patient would not have reported to hospital had he not been examined for an insurance policy, when it was suggested that he should seek further advice.

On Examination.—The patient appeared in good general health. In the neck there was a tumour in the right posterior triangle situated immediately behind the mid-point of the sternomastoid muscle; this was plainly visible and measured some 4 cm. vertically by 2·5 cm. horizontally. The swelling was hard on palpation and the surface was smooth except for one irregular nodule at its lower pole; the overlying skin was freely movable, but there was some deep attachment to the region of the carotid sheath. Two firm nodules a little less than 1 cm. in diameter could be felt above and separated from the main swelling.

Investigations.—(1) Blood-picture: Hb 13·8 g./100 ml., W.B.C. 7,200, differential white-cell count normal. (2) E.S.R. 2 mm. in first hour. (3) Radiographs of chest normal. (4) Radiography of neck showed a fine stippled calcification in the region of the main tumour.

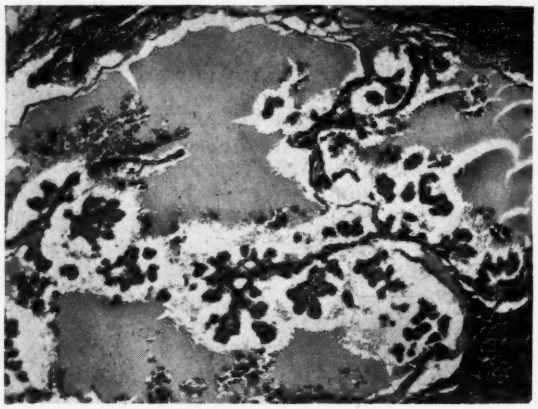

Fig. 417.—Low-power view of the main deposit. The histology is that of a papillary cystadenoma without evidence of malignancy. (× 36.)

At Operation.—On Jan. 23, under endotracheal gas-oxygen-ether anæsthesia, a horizontal incision was made over the swelling in the posterior triangle. Good exposure was obtained; the main tumour looked like a lymphatic gland and was closely related to the internal jugular vein. At its upper pole four other lymphatic glands were seen, two of these being the swellings previously felt while the other two were quite small. The tumour and related glands were not unduly vascular and were readily removed. The wound was closed in two layers without drainage.

Pathology.—The main tumour measured 3·0 by 2·3 by 1·3 cm., and on sectioning there was a well-marked fibrous capsule in which there were small areas of calcification and some lymphatic tissue. The cut surface of the tumour was rather spongy in appearance (*Fig.* 416); the two larger lymph-glands looked much the same, while the two smaller ones seemed normal. The histology of the main tumour was that of a papillary cystadenoma of the thyroid with little colloid content and some normal lymphatic tissue at the periphery. Similar changes were seen in the two larger lymphatic glands, with the additional feature of colloid-containing vesicles (*Fig.* 417). There was no evidence of malignant change in these sections and the remaining glands showed only a reactive sinus catarrh.

FURTHER PROGRESS.—The operation wound healed uneventfully and there was then some discussion as to the correct line of subsequent treatment. Pathological opinion favoured a diagnosis of papillary cystadenoma in aberrant thyroid tissue rather than metastases from a primary tumour in the thyroid gland. The neck was carefully re-examined, but the thyroid gland seemed quite normal. Surgical opinion differed on further management, and while some suggested masterly inactivity and observation others advocated exploration of the thyroid and hemithyroidectomy if any tumour was discovered. A study of the literature and especially of Crile's later paper left one in no doubt that hemithyroidectomy should be done, and this was accordingly advised.

FIG. 418.—Lateral aspect of right lobe of thyroid. The posterior surface of the lobe is on the left and the upper pole has been horizontally sectioned at the level where it was adherent to the larynx. The primary tumour was not detectable from the outside. (× 1½.)

SECOND OPERATION.—On Feb. 20 under endotracheal gas-oxygen-ether anæsthesia the thyroid gland was explored. A collar incision was made and the infrahyoid muscles were divided on the right side to allow easy exploration of this lobe. The whole gland was carefully examined and, apart from the fact that the right lobe seemed very slightly larger than the left, no abnormality was noted. The main thyroid arteries and veins were ligated and divided on the right side, the isthmus was divided to the left of the midline and the right lobe was removed, the recurrent laryngeal nerve being carefully defined and preserved. At this stage very slight adherence of the posteromedial aspect of the upper pole to the side of the larynx was noted. The wound was closed in layers with drainage to the right side.

PATHOLOGY.—The specimen consisted of the right lobe of the thyroid, measuring 5·0 by 2·8 by 1·6 cm., with part of the isthmus, the whole weighing 11·5 g. The surface of the specimen was normal (*Fig.* 418), but horizontal sectioning of the upper pole revealed a small tumour 1·8 by 1·2 by 0·6 cm. in size; this was situated posteriorly and was surrounded by a well-developed capsule up to 1 mm. thick and showing small foci of calcification (*Fig.* 419). The capsule was absent above and medially and here the pale greyish tissue of the tumour appeared to have spread beyond the gland.

FIG. 419.—Upper pole of gland showing cut surface after sectioning. The upper extremity of the gland has been left attached and turned over to show both cut surfaces. The small size of the primary tumour is well shown. (× 7/3.)

Histologically there was a cystic papillary structure with areas of dense fibrosis and calcification (*Figs.* 420, 421) resembling the glands previously removed from the neck. Here, however, there was clear evidence of infiltration and there was no doubt that this tumour was a papillary adenocarcinoma and that it was the source of the seemingly innocent deposits previously removed.

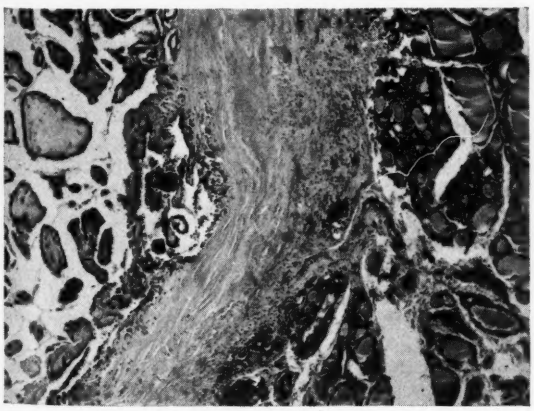

FIG. 420.—Low-power view of the edge of the primary tumour. Tumour on left, capsule in the centre, and normal gland on the right. (× 33·3.)

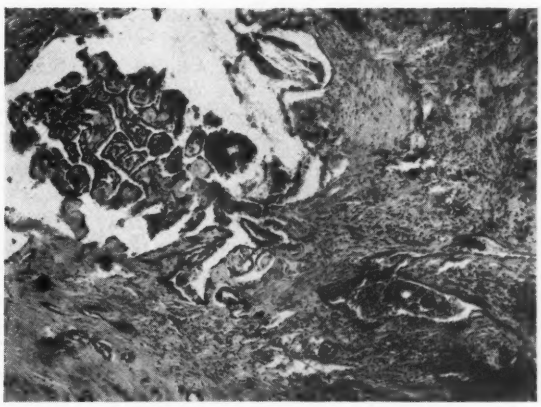

FIG. 421.—Higher-power view of the primary tumour, showing infiltration of the capsule (lower right) at the posterior aspect of the tumour. (× 83.)

SUBSEQUENT PROGRESS.—Following the second operation the patient's voice was hoarse and the right vocal cord was found to be inactive. The voice became normal again in a few weeks and the vocal cord was seen to be active.

A course of radiotherapy was given from March 15 to April 13, a total dosage of 3,000 r being delivered to the thyroid bed and the right anterior compartment of the neck.

The patient has been seen at three-monthly intervals since his discharge from hospital and has remained in good health. Periodic polarographic examination of the blood has been done and it is interesting to note that a recording of 12 micro-amps before the second operation had fallen to 6 micro-amps on Nov. 16 and was 8 micro-amps when last done on March 8, 1949. Polarographic examination was introduced by Brdička and an account of the principles and method of this examination was published by Robinson (1948), and by Robinson and Warren (1939). It remains to be seen whether periodic examinations of the blood by this method will provide a sensitive index of progress and of the development of metastases in cases of this type.

DISCUSSION

Papillary adenocarcinoma of the thyroid is frequently of very slow growth and low-grade malignancy and these remarks apply with similar force to the secondary deposits which so commonly manifest themselves before the primary tumour. Crile (1947) has published 21 cases of so-called 'lateral aberrant thyroid', in every one of which a primary tumour was discovered in the same side of the thyroid gland. Warren and Feldman (1949) have similarly published 49 cases where there was associated thyroid carcinoma, while in a further 8 cases of theirs the thyroid gland was never removed and the presence of a primary tumour was neither established nor disproved. When one bears in mind the fact that the thyroid tumour was never suspected clinically in 15 per cent of the cases where primary thyroid carcinoma was eventually proven, it would seem highly probable that there was a cancer in the thyroid of these other eight cases. The notoriously slow growth of both the primary and the secondary tumours makes it difficult to disprove the condition merely because of five or ten years of apparent freedom from trouble, for the lateral cervical swelling has been present for an average period of four years before treatment in most cases, and only 3 out of Crile's 21 cases had died during a follow-up varying between five and twenty-one years. Crile has a patient alive and well twenty-seven years after the appearance of lateral cervical tumours despite the fact that this patient refused all treatment, while the author has a patient under observation who is alive and well twenty-six years after the appearance of the lateral swellings. The latter patient was treated with deep X rays thirteen years ago and he still has multiple tender glands palpable on the affected side of his neck.

The tendency of these tumours and their lymphatic-gland metastases to undergo calcification may give rise to confusion and the X-ray shadows may lead to an unjustifiable diagnosis of calcified tuberculous glands.

The primary tumour is frequently situated on the posterior and medial aspect of the lateral lobes of the gland, a fact which is stressed by Crile and illustrated by the case reported here.

While there can be little argument about the advisability of hemithyroidectomy even when the gland seems normal to the naked eye, there is some difference of opinion over the management of the lateral deposits. King and Pemberton (1942) recommend radical block dissection in every case. Lahey supports this view, although he persists in the belief that the lateral deposits arise from the ultimobranchial bodies and he explains the presence of a tumour in the thyroid itself either by metastasis from the lateral tumour (that is, spread contrary to the normal lymphatic flow) or by the independent presence of other ultimobranchial tissue in the thyroid gland itself.

Crile and Warren and Feldman are of the opinion that block dissection is unnecessary and that hemithyroidectomy with simple dissection of the lymphatic glands on the same side is adequate. This was the course followed in the present case. The radiosensitivity of these papillary adenocarcinomata seems unquestionable and it is the practice at St. Bartholomew's Hospital to irradiate the neck post-operatively. Lahey advocates similar post-operative radiotherapy, but Graham and McWhirter (1947) are doubtful of its value in cases of papillary adenocarcinoma.

SUMMARY

A case is reported of occult papillary adenocarcinoma of the thyroid gland with cervical metastases initially diagnosed as lateral aberrant thyroid tissue.

The importance of exploring the thyroid gland and of performing hemithyroidectomy in *every case* is well shown by the present case and by many others reported in the literature.

The notoriously slow development of these tumours and several other misleading features are stressed.

Current views on the treatment of the cervical lymph-glands are discussed and the value of radiotherapy is considered.

I am much indebted to Dr. John W. Whittick for the pathological reports on this case; to Dr. George Cunningham for the photomicrographs, and to the photographic department of St. Bartholomew's Hospital for the pictures of the thyroid gland. Finally, I must thank Mr. E. G. Tuckwell for permission to treat this patient, who was admitted under his care.

REFERENCES

CRILE, G., jun. (1939), *Surg., Gynec. Obstet.*, **69**, 39.
— — (1947), *Ibid.*, **85**, 757.
GRAHAM, J. M., and MCWHIRTER, R. (1947), *Proc. R. Soc. Med.*, **40**, 669.
KING, W. L., and PEMBERTON, J. de J. (1942), *Surg., Gynec. Obstet.*, **74**, 991.
LAHEY, F. H., and FICARRA, B. J. (1946), *Ibid.*, **82**, 705.
ROBINSON, A. M. (1948), *Brit. J. Cancer*, **2**, 360.
— — and WARREN, F. L. (1949), *J. Path. Bact.*, **60**, 152.
WARREN, S., and FELDMAN, J. D. (1949), *Surg., Gynec. Obstet.*, **88**, 31.

SOLITARY KIDNEY

A REPORT OF THREE CASES

By V. S. HOWARTH

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THE incidence of this anomalous condition is given by Hinman (1935) as one in a thousand of four thousand autopsies. Macalpine (1949) states that its incidence has been variously estimated at one in fifteen hundred, and he quotes Guizzetti and Pariset as placing it at one in five hundred, but adds that he considers this latter figure to be too high.

Thomson-Walker (1936) collected 93 cases of death from uræmia or anuria commencing within the first few days after an operation on one kidney and found that there was no second kidney in 10 of the cases, and that the second kidney was "completely atrophied" in 8 cases. In over 19 per cent of cases the fatal result was therefore due to absence or atrophy of one kidney. The last-named author also notes the frequency of disease processes affecting solitary kidneys, calculus formation being especially frequent, while malignant growths, tuberculous disease, hydro- and pyonephrosis have been recorded. He also points out that congenital abnormalities of the genital organs, or other congenital abnormalities, should put the surgeon on his guard when dealing with a case of kidney disease, and quotes Ballowitz's record of a 70·8 per cent incidence of congenital abnormality of the genitalia associated with a congenital solitary kidney.

The recent occurrence of three examples of this anomaly illustrating some of the above features and the surgical importance attached thereto, therefore warrants a record.

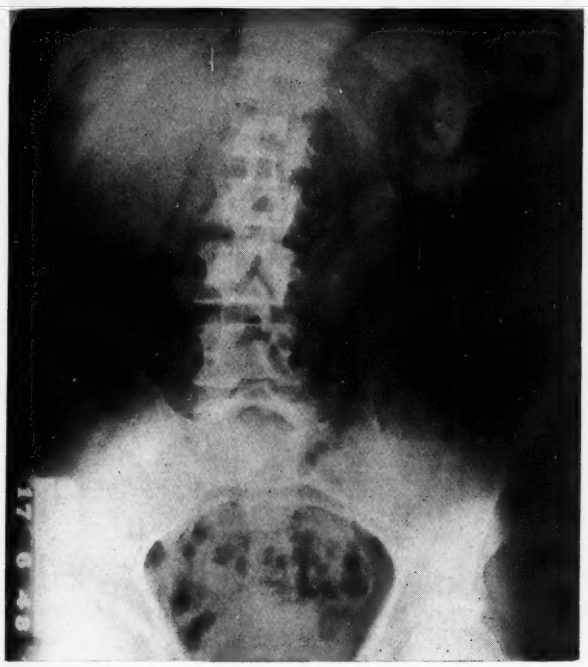

FIG. 422.—*Case* 1. An intravenous urogram. There is no kidney shadow or renal function to be seen on the right side. The left renal pelvis is anatomically normal and the kidney shadow is large.

FIG. 423.—*Case* 2. An intravenous urogram. Note the large renal shadow on the left side. There is no renal shadow or function to be seen on the right side.

CASE REPORTS

Case 1.—A male, aged 57, complained of pain in the back and right hypochondrium on and off for ten years. Ten years previously he had been admitted to hospital and investigations had been carried out to elucidate the nature of his symptoms and he had been informed on this occasion that his right kidney was "dead". The pain of which the patient complained was constantly present and was aggravated by exertion. He said that it had been getting worse for the past three to four months. There were no symptoms referable to the urinary system.

On abdominal examination a rounded smooth tumour was palpated in the right hypochondrium. It extended about 3 in. below the costal margin, moved with respiration, and could not be distinguished from the liver.

Graham's test was performed and found to be negative.

Cystoscopic examination was carried out and no abnormality was detected. On attempting to pass a catheter into the right ureter, an impassable obstruction was encountered just proximal to the ureteric orifice.

An intravenous urogram was obtained (*Fig.* 422).

Exploration of the right loin was carried out. The posterior abdominal wall was searched from the

diaphragm to the brim of the pelvis and no trace of a kidney or ureter could be found.

On palpation through the posterior parietal peritoneum a tongue-shaped process could be felt extending downwards from the right lobe of the liver to the right iliac fossa. It was assumed to be a Riedel's lobe.

The patient was seen recently and eight months after the operation he still complains of the same symptoms.

Case 2.—A female, aged 15, complained of recurrent attacks of pain localized to the right iliac fossa. The pain was associated with the menstrual flow. There had been no urinary symptoms.

FIG. 424.—*Case* 2. A retrograde pyelogram which demonstrates a bifid left renal pelvis.

Appendicectomy had been performed two years before with no relief, and gynæcological examination had led to a laparotomy being performed at which a bicornuate uterus had been discovered.

An intravenous urogram was obtained (*Fig.* 423).

Cystoscopic examination was carried out and the right half of the trigone was found to be absent and a ureteric orifice could not be discovered on this side. The left ureter was catheterized with ease and a left ascending urogram obtained (*Fig.* 424).

Case 3.—A male, aged 55, complained of an attack of painless hæmaturia twelve years previously, following which he had been free of symptoms up till three years ago, following which attacks had been present on and off up till the present. The hæmaturia was associated with exertion, and because of this fact the patient gave up his work and golf.

An intravenous urogram was obtained (*Fig.* 425).

A cystoscopic examination was carried out and a vesical calculus seen.

At this examination the left half of the trigone was noted to be absent. A left ureteric orifice could not be discovered and an unsuccessful attempt was made to catheterize the right ureter. Indigo carmine was excreted from the right ureteric orifice three and a half minutes after intravenous injection, but no dye could be seen coming from the left side.

Litholapaxy was performed.

SUMMARY

1. Three examples of congenital solitary kidney are reported.

2. There were associated congenital abnormalities present in two of the three cases recorded, *Case* 1 being associated with a Riedel's lobe and *Case* 2 with a bicornuate uterus and a contralateral bifid kidney pelvis.

FIG. 425.—*Case* 3. An intravenous urogram. Note the large renal shadow on the right side. There is no renal shadow or excretion of dye to be seen on the left side.

3. A vesical calculus was present in *Case* 3.

4. Exploration of the loin is necessary for an absolute diagnosis when, (*a*) the corresponding orifice of the ureter is normally developed in the bladder, but will not allow the passage of a catheter, and, (*b*) there is no renal function on the corresponding side shown by intravenous urography (*Case* 1).

I am indebted to Mr. J. W. S. Laidley and Mr. A. L. Carrodus, of the Department of Urology, Royal Prince Alfred Hospital, Sydney; and to the Department of Medical Artistry, The University of Sydney, for the photography.

REFERENCES

HINMAN, F. (1935), *Principles and Practice of Urology*, 460. Philadelphia and London: W. B. Saunders Co.

MACALPINE, J. B. (1949), *Cystoscopy and Urography*, 395. Bristol: John Wright & Sons.

THOMSON-WALKER, SIR JOHN (1936), *Surgical Diseases and Injuries of the Genito-urinary Organs*, 71. London: Cassell & Co.

RETROPERITONEAL CYSTIC ADENOMA

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BASSINI (1889) described a retroperitoneal cystic adenoma similar to a cystadenoma of the ovary, and Frank (1894) also observed such a tumour and attributed its histogenesis to an accessory ovary.

A tumour with these features is recorded in the following case report.

CASE REPORT

A female, aged 37, first noticed a swelling in her abdomen following a miscarriage fifteen months before being seen. The lump had slowly increased in size and was not associated with any pain or discomfort other than a dragging sensation which she referred to the left side of her abdomen. There had been no urinary symptoms or gastro-intestinal complaints. No menstrual irregularities had been noted. The patient had gained 2 st. in weight over the past two years.

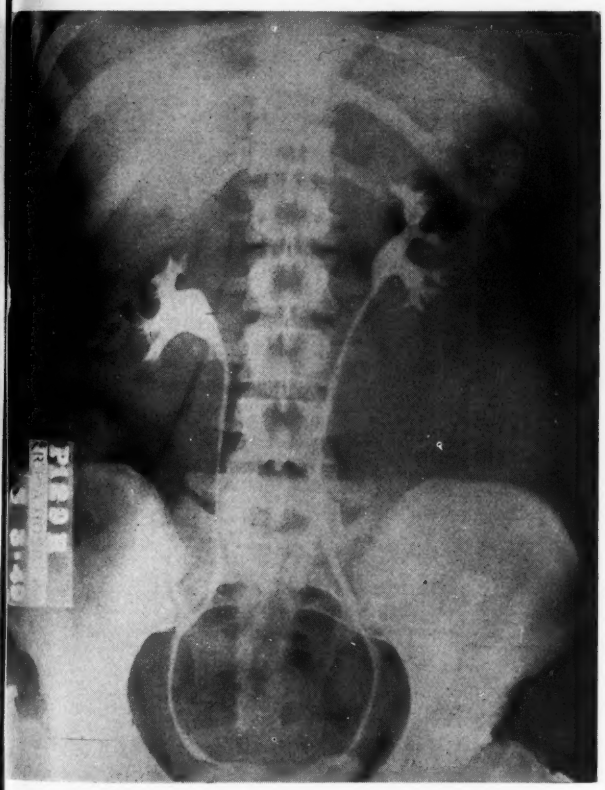


FIG. 426.—A bilateral retrograde pyelogram. Both kidney outlines can be seen to be normal in shape and position and the outline of the renal pelves does not show any abnormality. The left ureter has been displaced towards the midline by a soft tissue shadow which occupies the left flank, left iliac fossa, and overlies the lower pole of the left kidney.

ON EXAMINATION.—Inspection of the patient's abdomen disclosed an obvious swelling extending from the left costal margin almost to Poupart's ligament, round in shape, with maximum convexity at the level of the umbilicus. It did not transgress the midline. On palpation the tumour was firm, tensely cystic, and could be moved about an inch in all directions. It was possible to insinuate the fingers below its lower pole, but the upper pole receded beneath the costal margin. On bimanual palpation with one hand in the left lumbocostal triangle a sensation of ballottement could be obtained. On vaginal examination the uterus was found to be of normal size and no attachment of the tumour to the uterus or adnexa could be determined.

No abnormal constituents were detected on examination of the patient's urine and hæmatological examinations yielded normal findings. The blood-Wassermann reaction was negative as was the blood complement-fixation test for hydatid disease. The Casoni intradermal reaction for hydatid disease did not give a positive result. A plain radiograph of the abdomen showed a large well-defined mass occupying the left iliac fossa and extending up to the lower pole of the left kidney.

Retrograde pyelography was performed (*Fig.* 426).

AT OPERATION.—A transperitoneal approach was made to the tumour via a left paramedian incision. The descending colon was found to be stretched over the medial aspect of a large, tensely cystic structure which occupied the retroperitoneal tissues, extending upwards to overlie the lower pole of the left kidney and downwards into the left iliac fossa. The left ureter was displaced towards the midline by the cyst, which was removed without difficulty and without the discovery of any definite attachment.

THE SPECIMEN.—*See Figs.* 427, 428.

HISTOLOGY OF THE CYST.—*See Figs.* 429, 430.

DISCUSSION

Handfield-Jones (1924), in a classic account of retroperitoneal cysts, clearly recognizes the ample possibilities of remnants of the embryonic urogenital system to give rise to cysts in the retroperitoneal tissues, but points out that there is nothing conclusive as to their histology and that a dogmatic expression of opinion is not justified as to their histogenesis. Hansmann and Budd (1931) described 17 primary retroperitoneal tumours, and found the histology of these tumours similar to that of tumours arising in the adult urogenital apparatus. Donnelly (1946), reporting 95 cases and reviewing the literature, finds that " primary retroperitoneal tumours occur with sufficient frequency and that they should always be considered in the differential diagnosis of an unexplained abdominal mass ". Nichols (1947) finds that there are two general types of primary developmental retroperitoneal cysts, one type being derived from the Wolffian body or genital ridge and showing in its wall epithelial elements, occurring in women and occupying a lateral position, and the other composed of mesodermal or lymphatic elements occurring in any age group, in the midline or occupying a lateral position.

All writers on this subject have stressed the value of pyelography, either instrumental or descending, both as a diagnostic measure and in the planning of the subsequent operative approach, and the employment of this measure enabled an accurate pre-operative diagnosis to be made in the case herein reported.

FIG. 427.—The tumour prior to section. (× ½.)

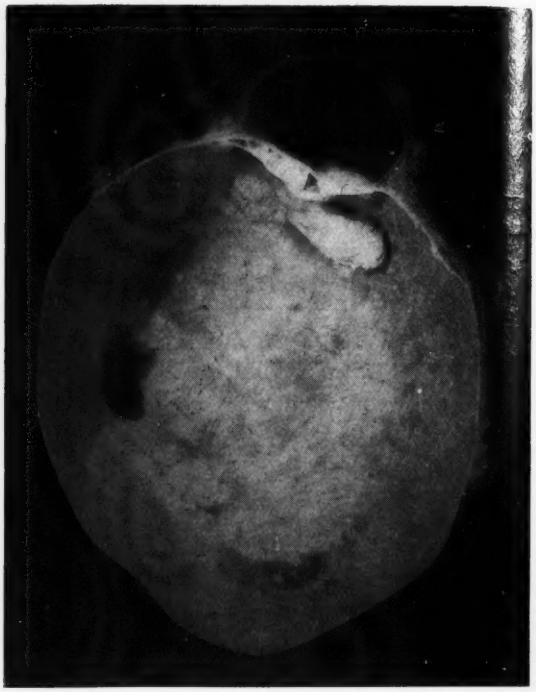


FIG. 428.—A coronal section of the tumour demonstrates it to be a bilocular cyst. Its lining is white and glistening and at one point a definite intracystic nodular projection can be seen and an incision into this portion demonstrates it to be composed of cystic spaces containing mucoid material. Microscopical sections are taken from this area. Both loculi contained chocolate-coloured fluid which shimmered with crystals. (× ½.)

SUMMARY

1. A retroperitoneal cyst with histology similar to that of some epithelial cysts of the ovary is reported.

2. Pyelography is invaluable in the diagnosis of retroperitoneal tumours.

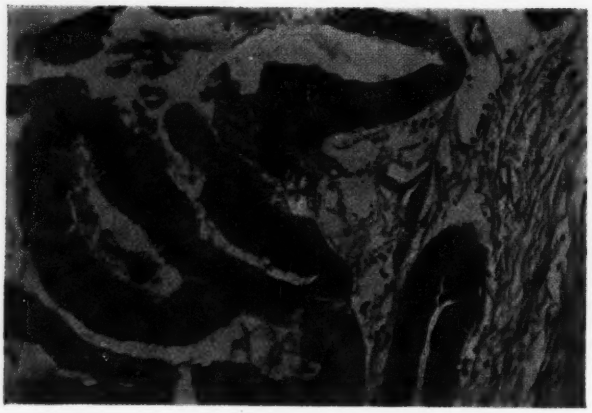


FIG. 429.—The epithelium is of a tall columnar type reminiscent of that seen in pseudomucinous ovarian cystomata. Epithelial-lined spaces can be seen lying in a fibrous tissue stroma beneath the epithelial lining of the cyst. The degree of epithelial activity and its darkly-staining nuclei would suggest a malignant, rather than a benign, tumour H. and E. (× 400.)

3. An origin from remnants of the genital ridge is suggested.

4. A short review of the literature is included.

I am indebted to Mr. F. W. Niesche, of the Department of Surgery, Royal Prince Alfred Hospital, Sydney, for permission to publish this case record; and to the Department of Medical Artistry, The University of Sydney, for the photography.

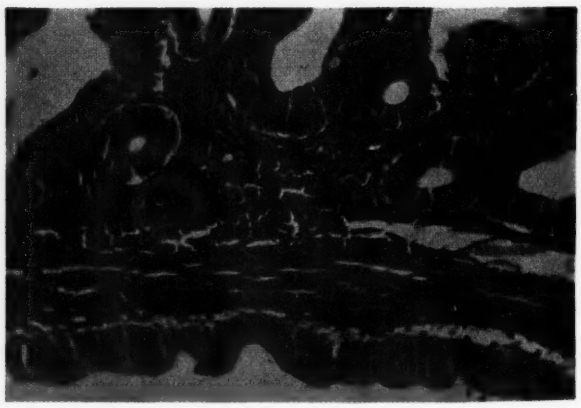


FIG. 430.—Southgate's stain for mucin. This section demonstrates the production of mucin by the cyst epithelium. (× 400.)

REFERENCES

BASSINI, E. (1889), quoted by DONNELLY.

DONNELLY, BERNARD A. (1946), *Surg. Gynec. Obstet.*, **83**, Dec. 705.

HANDFIELD-JONES, R. M. (1924), *Brit. J. Surg.*, **12**, 119.

HANSMANN, G. H., and BUDD, J. W. (1931), *Amer. J. Path.*, **7**, 631.

NICHOLS, H. MINOR (1947), *Ann. Surg.*, **126**, 340.

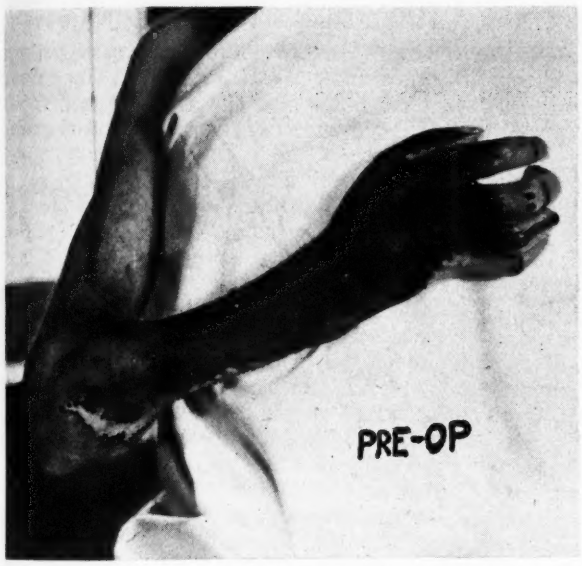
STEWART, LESLIE R. (1926), *Edinb. med. J.*, **33**, 441.

CASE REPORT ON PEDICLED NERVE-GRAFT*

By F. G. ST. CLAIR STRANGE, Canterbury

A preliminary communication describing an operation for nerve pedicle grafting was published in this journal in 1947 and the present paper describes the outcome of what was then a purely experimental operation performed on theoretical grounds.

To summarize the case in a few words, it may be stated that a 12-cm. gap in the right median nerve in the forearm was bridged in two stages by a graft from the ulnar nerve, which had a similar gap at the same level (*Figs.* 431, 432). The first step



Figs. 431, 432.—Anhidrosis, anæsthesia, and analgesia of the median and ulnar areas. There is a little radial and musculocutaneous overlap onto the thenar eminence, and an unusually small autonomous ulnar distribution. In these and the following photographs a quinazirin sweating test has been performed, loss to light touch is outlined by a continuous line and to superficial prick by a dotted line. (The forearm scars are also outlined by a finely-dotted line in some of the illustrations.)

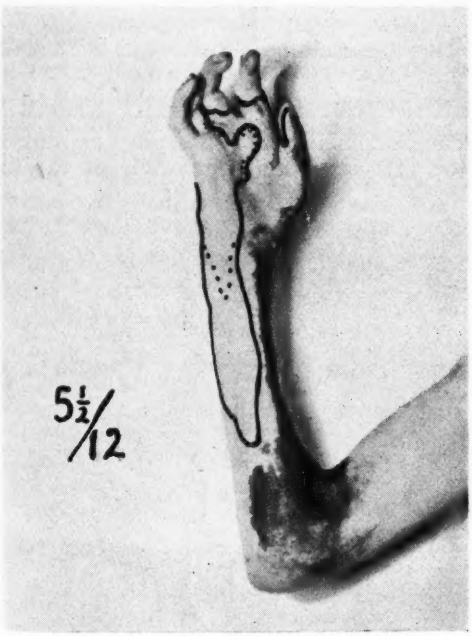


Fig. 433.

Fig. 434.

* Based on a paper read at the combined meeting of the British, Canadian, and American Orthopædic Associations at Quebec, June, 1948.

Fig. 435.

FIGS. 433–435.—Successive progress illustrations at 5½, 7, and 16 months after the first operation.

consisted of suturing the proximal faces of the two nerves together and dividing the ulnar nerve high up, at the same time retaining its blood-supply. At the second stage the ulnar graft was swung down and sutured to the distal stump of the median, now gaining its blood-supply across the first suture line. This operation was planned to avoid any period of ischæmia, thus preserving the viability of the Schwann cells and allowing axons to traverse the graft in ideal circumstances.

The end-result has been most satisfactory on the sensory side, fulfilling Seddon's stringent standards (1947), namely, that for a graft to be considered a success it must achieve a result at least as good as the best results of direct suture.

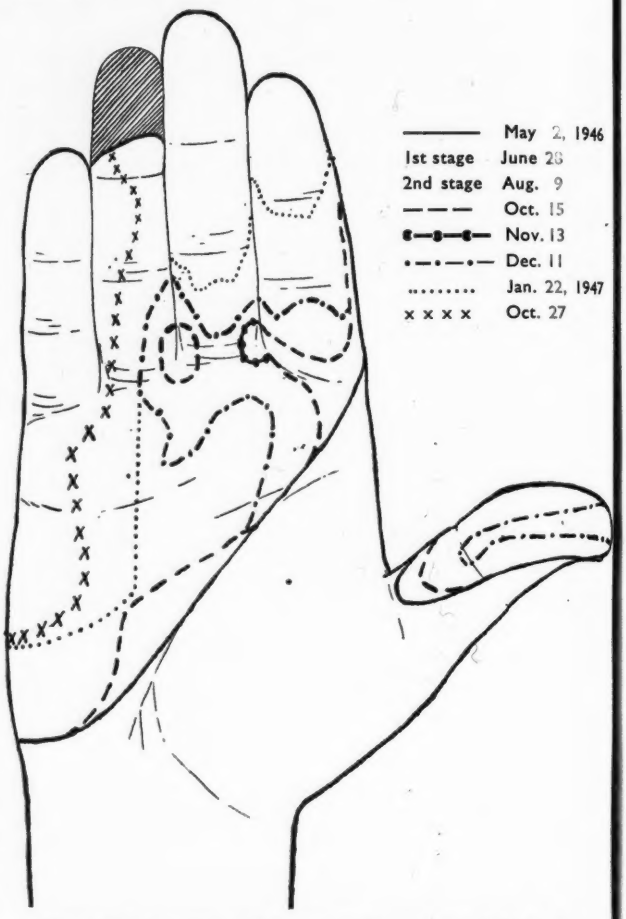
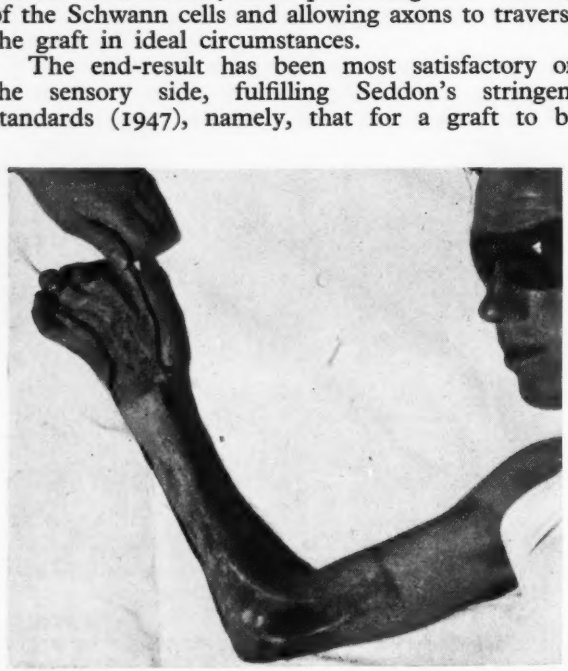
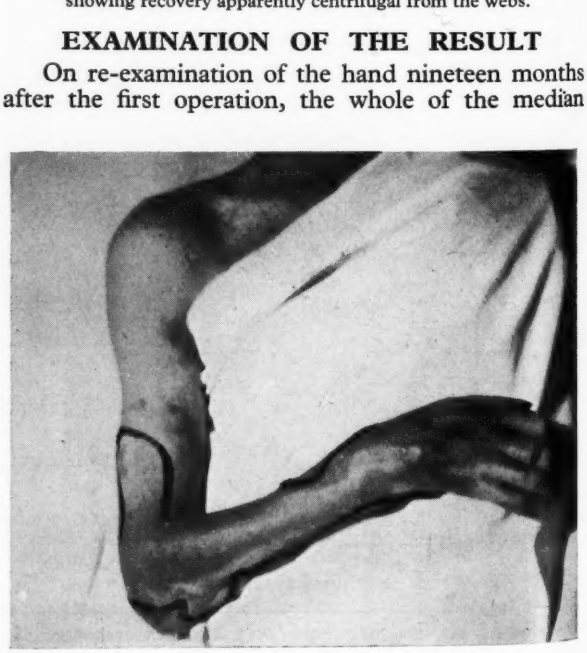


FIG. 436.—A summary of the return of response to light touch, showing recovery apparently centrifugal from the webs.

EXAMINATION OF THE RESULT

On re-examination of the hand nineteen months after the first operation, the whole of the median





FIGS. 437, 438.—Sweat test and sensory chart of condition at radial block, 19 months after operation. The median nerve alone continues to conduct impulses.

area had become re-innervated (*Fig.* 435). Response to light touch and superficial prick were almost normal throughout, there being only a small patch of hypoæsthesia, 1·5 cm. in diameter, in the centre of the palm, and slight over-reaction to prick in the palm, though not in the pulps of the fingers. Two-point discrimination was nearly always right at 1 cm. separation of the divider points.

Deep pressure is recognized throughout without pressure pain, and localization is accurate in the thumb and index. In the third finger it is sometimes referred to the base of the fourth.

Postural sense is present and accurate in all median-supplied joints to medium coarse movements. Temperature sense is present and accurate, though still a little slow in appreciation. Vibration sense is present in the palm and thumb, but has not re-appeared in the fingers.

Motor function in the thenar muscles has not recovered to any perceptible amount, possibly due to their having an ulnar innervation, or possibly to the delay, something like two and a half years from the time of injury, before axons could have reached them again.

The skin has practically lost its shine and the finger pulps are thickening up again, though the nails are still a little curved, but no longer ridged.

Tendon transplants have since been done to give flexion to the fingers and opposition to the thumb.

The hand as a whole has returned into the patient's ken, and now restores him to the position of a two-handed individual. He can turn the pages of a newspaper and even get a match out of a box and strike a light.

On studying this case for presentation, recovery almost seemed to be centrifugal from the points of radial overlap in the first three webs (*Figs.* 433, 434, 436). I wondered if this might possibly have spread over the whole hand, and therefore performed a radial block (Highet, 1942). This was completely successful, producing paralysis, anhidrosis, and complete anæsthesia of the whole radial distribution, while the median area retained its sensation (*Figs.* 437, 438). This conclusively proves that all the sensory recovery in the median area has come through the graft. The rate of recovery through the graft is shown in *Fig.* 439.

DISCUSSION

This operation may have a very limited applicability, but the case history does show that it will work. The principle of moving a nerve-graft to its new site by a rational pedicle method, so as to retain its blood-supply, is established. It should therefore be possible to be certain that failure will not be due to ischæmic necrosis or collagenization of the graft where this method is capable of being used, as may occasionally occur in the many automobile and industrial accidents of peace time.

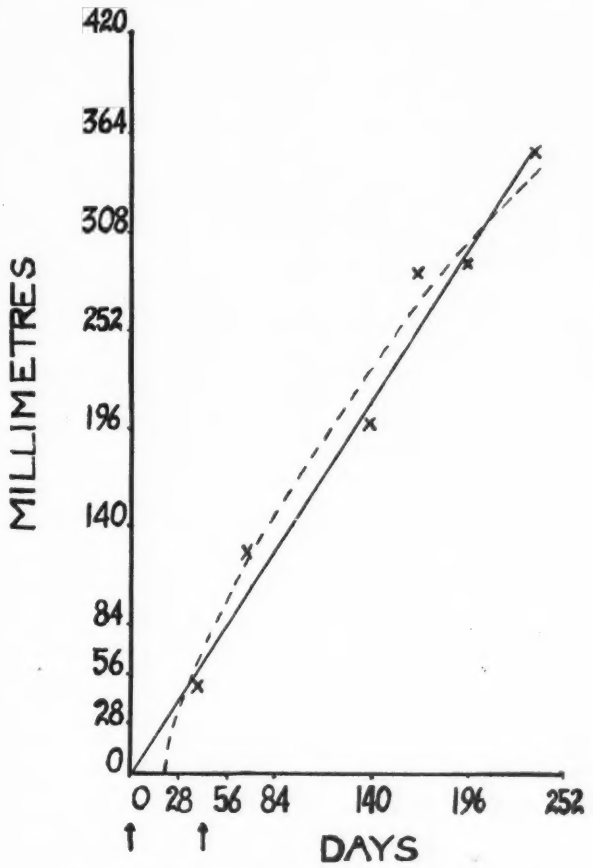


FIG. 439.—Recovery rate through the graft as estimated by observing the advance of Tinel's sign. Overall average 1.51 mm. per day.

SUMMARY

An account is given of the successful outcome of a previously described rational pedicled nerve autograft.

REFERENCES

HIGHET, W. B. (1942), *J. Neurol. Psychiat.*, No. 3 and 4, 101.
SEDDON, H. J. (1947), *Brit. J. Surg.*, **35**, 151.
STRANGE, F. G. ST. C. (1947), *Ibid.*, **34**, 423.

THE ANATOMY OF THE BLOOD-VESSELS IN THE REGION OF THE PANCREAS

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OPERATIONS on the pancreas are being undertaken with increasing frequency, and the vascular arrangements in and around the gland have now considerable surgical importance.

The arteries have been exhaustively studied by many observers, and an accurate knowledge of their pattern has been gradually obtained. On the other hand, accounts of the venous drainage of the pancreas, and of the tributaries of the portal vein and their principal variations, are inadequate.

The present study was undertaken to amplify the standard descriptions and if possible to provide a guide to surgeons interested in the surgery of the pancreas and the duodenum.

The earliest accounts of the pancreatic arteries were lacking in detail; arcades between the pancreatic arteries arising from the cœliac axis and those from the superior mesenteric artery were not recognized until Haller described anterior and posterior arches in 1748; later Verneuil (1851) and Wiart (1899) modified Haller's original description. Brun (1913) repeated Haller's original description in more modern terminolgy, and from time to time several authors have added various details. Thus in his study of the supraduodenal artery, Wilkie (1911) reported a vessel, now identified as the posterior superior pancreatico-duodenal branch, which sprang from the gastroduodenal artery; later this vessel was the subject of more complete investigation by Flint (1922), Yule (1927), and Edwards (1941).

Petren undertook a very full review of the literature in 1929. His account was illustrated by excellent drawings and, with its clear nomenclature, provided an accurate description of the vessels of the head of the pancreas, but he did not discuss the vasculature of the neck and body.

Wharton (1932) described a branch of the superior mesenteric artery—the inferior pancreatic—which coursed along the lower border of the pancreas, and a contribution of three to five pancreatic branches from the splenic artery. He also identified anterior and posterior pancreatico-duodenal veins, a group of pancreatic tributaries of the splenic vein, and several other inconstant tributaries which joined the portal vein from the head of the gland.

Kirk (1932) described a "supreme pancreatic artery" which appeared to supply the neck and body of the gland, and which took origin from the cœliac axis or from the splenic artery; and also a middle pancreatic arterial arcade buried in the substance of the head of the gland. In his account of the veins, he drew attention to an inferior pancreatic vein originally described by Tonkoff (1903) and a vein draining the neck of the pancreas which he named the "vena colli pancreatis".

Pierson (1943) reviewed the arterial blood-supply of the pancreas in 50 dissections, but dismissed the veins briefly as accompanying and superficial to the arteries.

Descomps and de Lalaubie (1912), in a series of dissections of the mesenteric veins, made some valuable observations on the venous drainage of the pancreas and on the tributaries of the superior mesenteric, portal, and splenic veins; they also described variations in the colic veins and illustrated the vena colli pancreatis, although it was not mentioned in their text.

MATERIAL AND METHODS

In the present review, 50 specimens—27 dissections and 23 injection-corrosion preparations—were studied in detail, the material being obtained at autopsy. In most specimens the liver, bile-ducts, stomach, duodenum, pancreas, and spleen were removed in one piece by the pathologist, and the dissection was continued after partial fixation in Jore's and Kaiserling's fluids. In the remaining specimens the liver and spleen had been removed separately.

In the dissected specimens the arteries and veins were traced without preliminary injections of the vessels; even the finer radicles were followed without difficulty, except in the region of the bile-ducts where post-mortem staining rendered recognition difficult.

In the injection-corrosion preparations the technique described by Morison (1923) was employed, the veins being injected with celloidin in a strength varying from 5 to 10 per cent. In four specimens neoprene (latex) was used as the injection medium; the technique followed was that described by Trueta, Barclay, Daniel, Franklin, and Pritchard (1947). The pressure used for the injection and the amount injected varied in different specimens owing to the impossibility of obtaining a closed circuit, and to a tendency to leakage from rupture of the finer unsupported vessels. Pressures of 100–300 mm. Hg were required to obtain visible fillings of the superficial radicles.

RESULTS

THE ARTERIES OF THE PANCREAS AND DUODENUM

The arterial distribution is conveniently considered in relation to (1) the head of the pancreas, and (2) the neck and body.

1. The Head of the Pancreas.—The head of the pancreas invariably derived its arterial blood-supply from anterior and posterior arcades formed by anterior and posterior pancreatico-duodenal arteries.

In every case the anterior vessel was a terminal branch of the gastroduodenal artery, which coursed over the head of the gland to reach the duodeno-pancreatic sulcus at the inferior border of the pancreas and then turned medially and backwards in the groove, until it reached the posterior aspect of the gland. An anastomosis was then established behind the uncinate process with an anterior inferior pancreatico-duodenal branch of the superior mesenteric artery (*Fig.* 440).

In every dissection both the inferior pancreatico-duodenal arteries were branches of the superior mesenteric artery. In 17 specimens they shared a common trunk with the first jejunal branch, and in 7 they originated in a common trunk situated proximal to the first jejunal branch. In 3 cases the anterior and posterior vessels had separate origins from the superior mesenteric artery (*Fig.* 440).

2. Neck and Body of the Pancreas.—The blood-supply of the neck and body of the pancreas

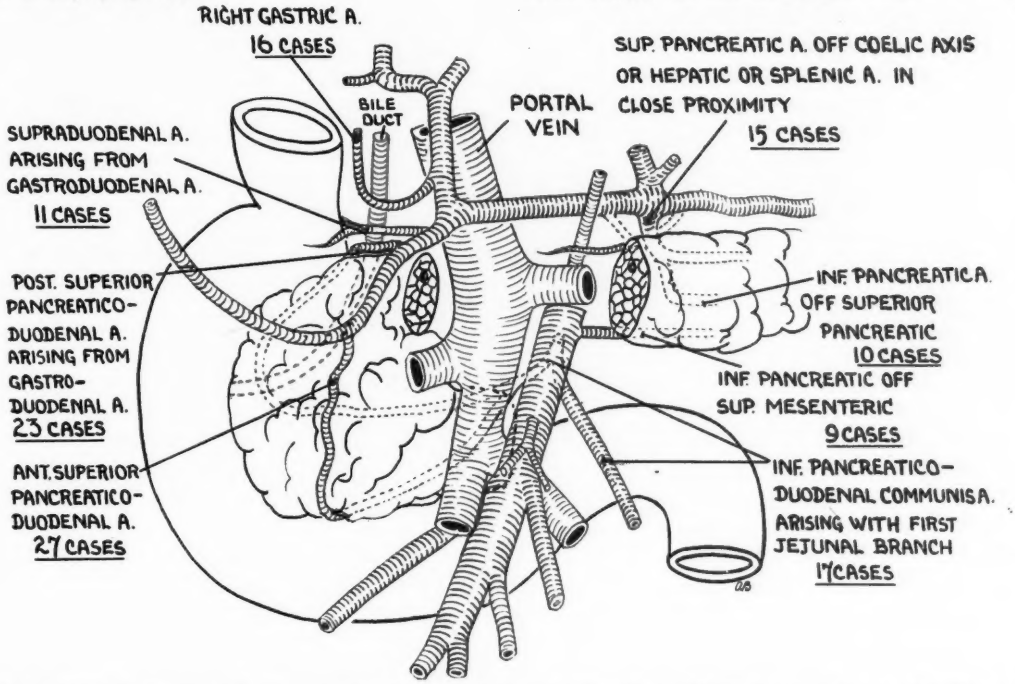
RIGHT GASTRIC A.
16 CASES
BILE DUCT
PORTAL VEIN
SUP. PANCREATIC A. OFF COELIC AXIS OR HEPATIC OR SPLENIC A. IN CLOSE PROXIMITY
15 CASES
SUPRADUODENAL A. ARISING FROM GASTRODUODENAL A.
11 CASES
POST. SUPERIOR PANCREATICO-DUODENAL A. ARISING FROM GASTRO-DUODENAL A.
23 CASES
ANT. SUPERIOR PANCREATICO-DUODENAL A.
27 CASES
INF. PANCREATIC A. OFF SUPERIOR PANCREATIC
10 CASES
INF. PANCREATIC OFF SUP. MESENTERIC
9 CASES
INF. PANCREATICO-DUODENAL COMMUNIS A. ARISING WITH FIRST JEJUNAL BRANCH
17 CASES

FIG. 440.—Arterial supply of pancreas, showing most frequently occurring distribution of vessels in 28 dissections.

The posterior superior pancreatico-duodenal artery was observed in 25 dissections. In all but two it arose from the gastroduodenal branch of the hepatic artery, 1·5 cm. distal to its origin. It coursed backwards over the upper border of the gland, and invariably in front of the common bile-duct, to reach the posterior pancreatico-duodenal sulcus; and after a short distance it left the sulcus and passed medially across the back of the pancreatic head, to an anastomosis with the posterior inferior pancreatico-duodenal artery (*Figs.* 441, 442).

In two exceptions, the posterior superior pancreatico-duodenal artery originated from an accesory hepatic branch of the superior mesenteric artery. In each case this anomalous hepatic artery passed behind the superior mesenteric vein, and ascended to the liver between the portal vein and the common bile-duct, with the normal hepatic artery in front of it. The posterior superior pancreatico-duodenal branch arose immediately distal to the origin of the accessory hepatic vessel, and its terminal branches fanned out to supply the whole of the posterior aspect of the head.

In one specimen the posterior inferior pancreatico-duodenal artery was absent, and there was no posterior anastomosis; but arcades were present between the individual branches of the posterior superior vessel.

is normally derived from inferior and superior pancreatic arteries. The inferior pancreatic artery was identified in 24 dissections. In 9 it arose from the left aspect of the superior mesenteric artery below the neck of the pancreas, and, passing to the left, gradually sank into the substance of the gland. In 10 specimens it was a branch of the superior pancreatic artery, and descended behind the neck of the pancreas and the splenic vein, towards the inferior border of the gland. In 4 cases it sprang from the anterior superior pancreatico-duodenal artery, passed medially on the anterior surface of the head and then on the lower border of the neck and the body, superficial to the superior mesenteric vein. In one specimen it pursued a similar course from an origin in the right gastro-epiploic artery.

A subsidiary arterial arcade between a medial branch of the anterior superior pancreatico-duodenal artery and a lateral branch of the inferior pancreatic artery was noted 11 times.

A superior pancreatic artery was found in 15 cases. In most instances it arose as a short trunk from the cœliac axis directly (3 cases), hepatic artery (6 cases), or splenic artery (6 cases). It descended to reach the body of the pancreas superior to the splenic vein, and then divided into right and left branches for the neck and body of the gland respectively. In 3 dissections the superior pancreatic was a branch of the superior mesenteric

artery, arising close to the aorta; it then arched upwards and forwards over the splenic vein, to reach the gland. Other origins encountered were from the inferior pancreatic artery (2 cases), the

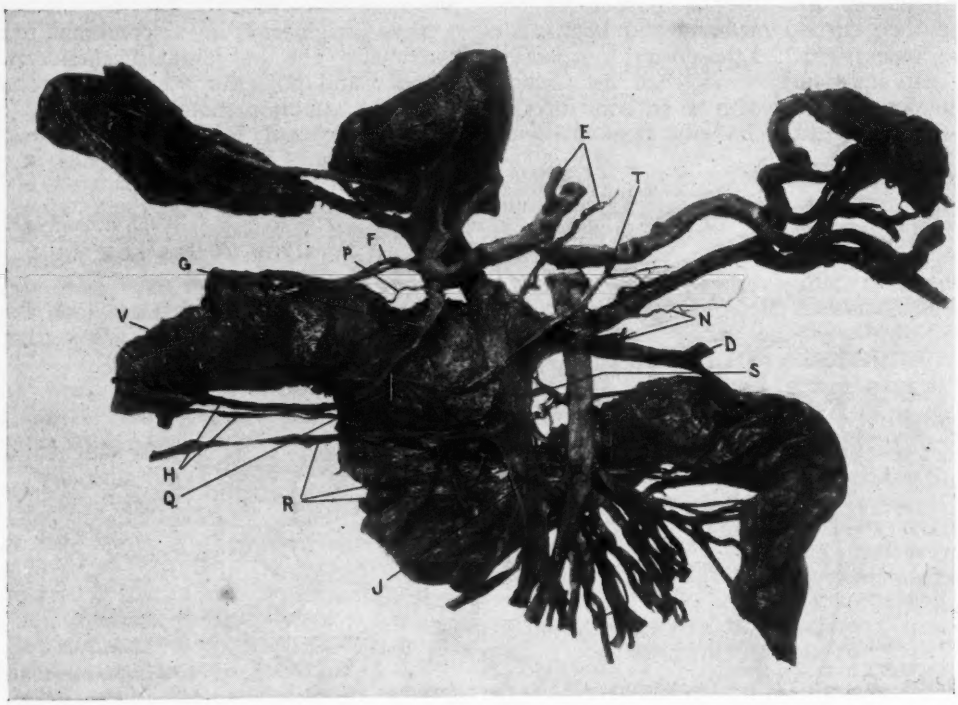

Fig. 441.—Anterior aspect of dissected specimen. D, Inferior mesenteric vein; E, Left gastric artery and vein; F, Right gastric artery and vein; H, Right gastro-epiploic artery and vein; J, Anterior inferior pancreatico-duodenal vein; N, Inferior pancreatic artery and vein; G, Posterior superior pancreatico-duodenal artery arising from gastroduodenal artery; P, Supraduodenal artery; Q, Anterior superior pancreatico-duodenal artery with anastomosis with superior pancreatic artery; R, Colic veins; S, Inferior pancreatico-duodenal artery; T, Superior pancreatic artery; V, Pyloric antrum reflected to right.

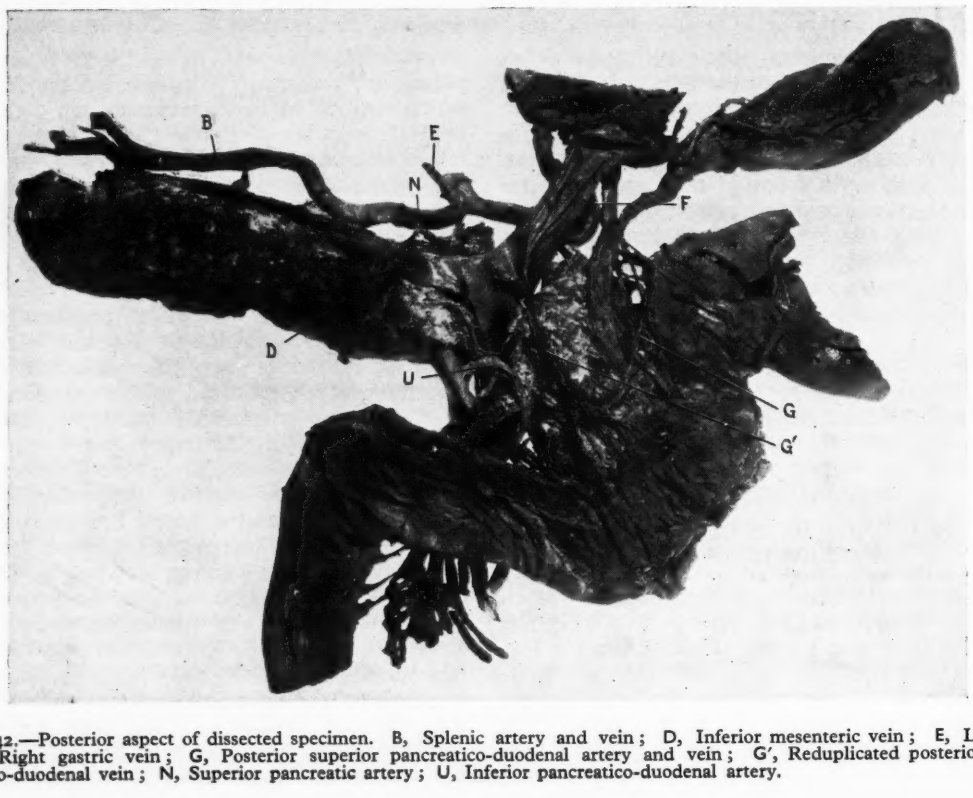

FIG. 442.—Posterior aspect of dissected specimen. B, Splenic artery and vein; D, Inferior mesenteric vein; E, Left gastric vein; F, Right gastric vein; G, Posterior superior pancreatico-duodenal artery and vein; G', Reduplicated posterior superior pancreatico-duodenal vein; N, Superior pancreatic artery; U, Inferior pancreatico-duodenal artery.

posterior superior pancreatico-duodenal artery (1 case), and the gastroduodenal artery (1 case); the vessels then coursed to the left, along the upper border of the neck and the body of the pancreas. In 5 dissections the superior pancreatic artery was not present. The body and tail of the pancreas also derived a varying number of short trunks from the splenic artery, which sank at once into the substance of the gland (*Fig.* 441).

The remarkably constant arrangement of the cœliac axis and the superior mesenteric artery in these dissections is worth comment. In 1 instance, however, the hepatic artery arose from the superior mesenteric and not from the cœliac trunk. Partly buried in the gland substance, it crossed the posterior aspect of the head of the pancreas and entered the free edge of the lesser omentum behind the portal vein, reaching its normal position by passing between the common bile-duct and the portal vein. An accessory hepatic branch from the superior mesenteric artery was noted on 3 occasions. It coursed posterior to the head of the pancreas, and in the margin of the lesser omentum it lay behind the normal hepatic artery, between the portal vein and the common bile-duct. An additional hepatic branch from the cœliac axis was found in 1 dissection; it lay behind the normal hepatic branch, reaching this position from behind the portal vein. The cystic artery was derived from the accessory hepatic vessels in 3 cases; in the fourth, the cystic artery could not be identified.

Summary.—

1. Anterior and posterior arterial arcades to the head of the pancreas are always present.

2. An inferior pancreatic artery is almost constant (24 of 27 dissections).

3. A superior pancreatic artery is frequent (22 of 27 dissections).

4. An accessory anterior arcade between the superior pancreatico-duodenal and inferior pancreatic arteries is an occasional finding.

5. The body and tail derive a variable number of small arteries from the splenic artery.

THE VENOUS ARRANGEMENTS IN AND ABOUT THE PANCREAS

The neck of the pancreas is the site of confluence of the entire venous drainage of the portal circulation. At that level the superior mesenteric and splenic veins unite to form the portal vein, which then sweeps up behind the neck of the pancreas towards the porta hepatis.

The portal vein was found to be constant in its formation; in all 50 specimens the only variations were in its smaller tributaries.

The superior mesenteric vein was usually formed by the confluence of two trunks. The right trunk, lying on the right of the superior mesenteric artery, received tributaries from the lower jejunum, ileum, and ascending colon. The left trunk received blood from the upper jejunum by several tributaries, and passed behind the superior mesenteric artery to join its fellow at a variable angle (18 of 27 dissections; *Fig.* 443). In 9 specimens, however, the left trunk crossed anterior to the artery and joined the right trunk obliquely at its junction with the splenic vein below the neck of the pancreas; the artery appeared between the two veins, and then pursued its usual course (*Fig.* 444).

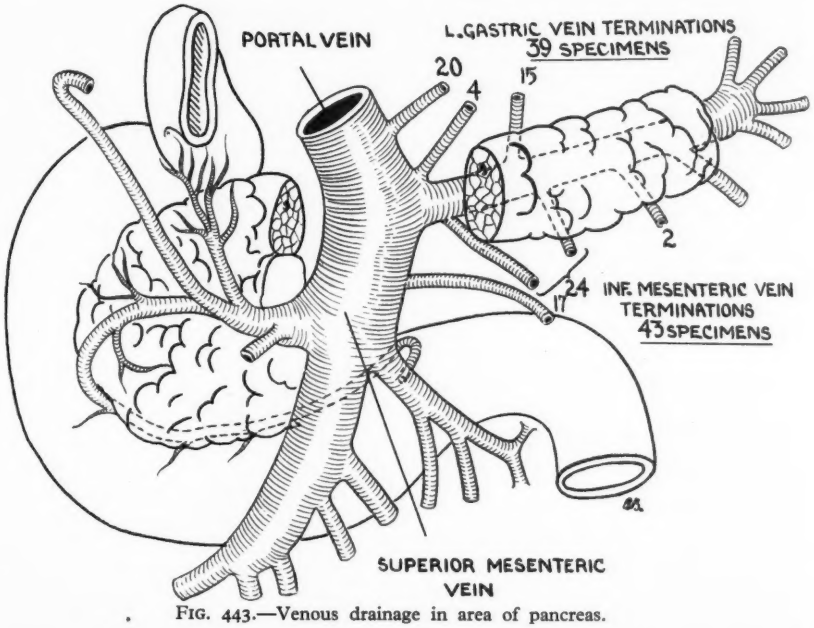

FIG. 443.—Venous drainage in area of pancreas.

The splenic vein was also relatively constant in its course, and joined the superior mesenteric vein behind the neck of the pancreas. Its main tributaries were the left gastric and the inferior mesenteric veins.

The left gastric vein ended variously in the last 2 cm. of the splenic vein (15 cases), the first 2 cm. of the portal vein (20 cases), or in the splenoportal junction (4 cases); it passed in front of the hepatic or the splenic artery, and then backwards behind the body of the pancreas, to the posteromedial aspect of the portal vein, or to the upper border of the splenic vein (*Figs.* 441–443, 445, 446).

The inferior mesenteric vein was a large trunk ending in the splenic vein (26 cases) or in the superior mesenteric vein (17 cases). The point of junction was usually within 3 cm. of the splenic termination, but in 2 dissections it was situated 10 cm. from the end of the splenic vein. When the inferior mesenteric joined the superior mesenteric system, it entered the main trunk on its medial aspect, superficial to the superior mesenteric artery (*Figs.* 441-443, 445, 446).

The right gastro-epiploic vein proceeded along the greater curvature of the stomach between the layers of the gastrocolic omentum, and entered the right extremity of the transverse mesocolon, in which it arched downwards and to the left through three-quarters of a circle to the head of the pancreas, and ended by joining the anterolateral aspect of the superior mesenteric vein, immediately below the neck of the pancreas (*Fig.* 447). In its course along the greater curvature it received tributaries

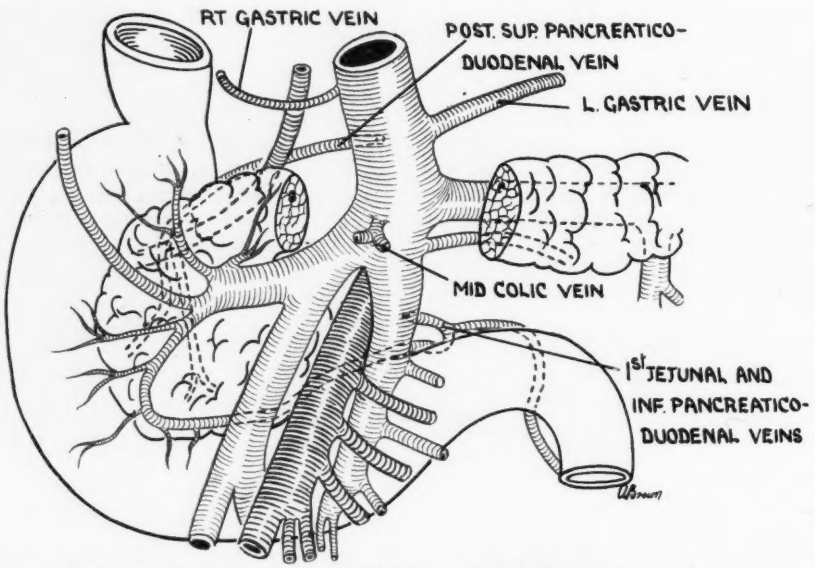

FIG. 444.—Formation of portal vein showing high union of the two limbs of the superior mesenteric vein.

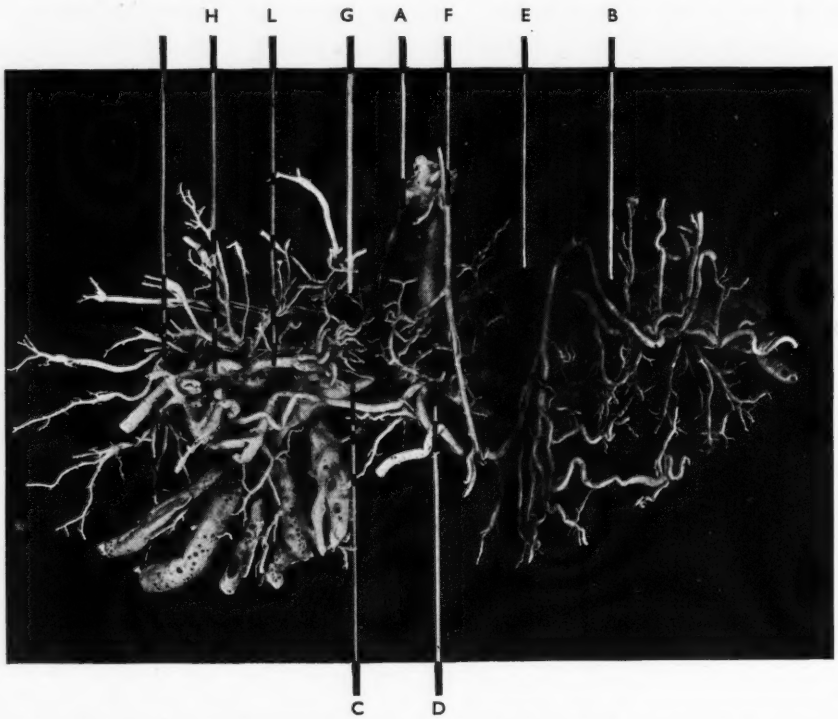

FIG. 445.—Injection-corrosion preparation from anterior aspect. A, Portal vein; B, Splenic vein; C, Superior mesenteric vein; D, Inferior mesenteric vein; E, Left gastric vein; F, Right gastric vein; G, Posterior superior pancreatico-duodenal vein; H, Right gastro-epiploic vein; I, Anterior superior pancreatico-duodenal vein; J, Anterior inferior pancreatico-duodenal vein; K, Gastrocolic trunk; L, Subpyloric vein; M, Posterior inferior pancreatico-duodenal vein; N, Inferior pancreatic vein; O, Colic vein.

from the stomach and omentum, and after leaving the omentum, in the last few centimetres of its course, from the pylorus and the first part of the duodenum (the subpyloric veins). Near its termination the gastro-epiploic vein was joined in all cases by the anterior superior pancreatico-duodenal and one or more colic veins. This arrangement is so constant that this part of the vein has been called the gastrocolic trunk (Descomps and de Lalaubie, 1912) (*Figs.* 441, 447, 448).

In our dissections the main venous drainage of the head of the pancreas was found to correspond closely to the description by Petren (1929), and his nomenclature has been followed. The upper and anterior part of the head was drained by the anterior superior pancreatico-duodenal vein formed in the pancreatico-duodenal sulcus near the end of the second part of the duodenum.

This vein passed upwards and, leaving the sulcus, turned medially to join the gastrocolic trunk. In

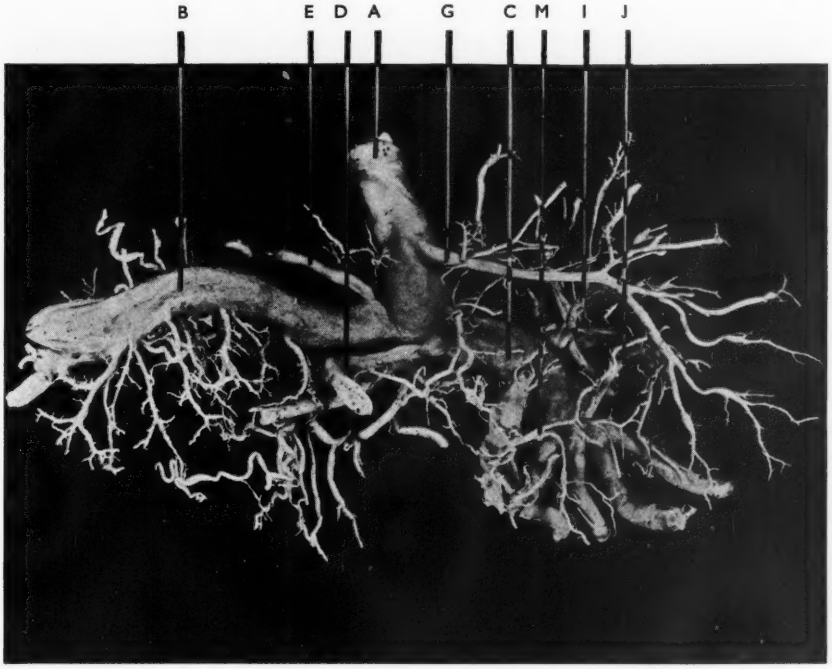

FIG. 446.—Posterior aspect of injection-corrosion preparation seen in *Fig.* 445. (For key to vessels, *see Fig.* 445.)

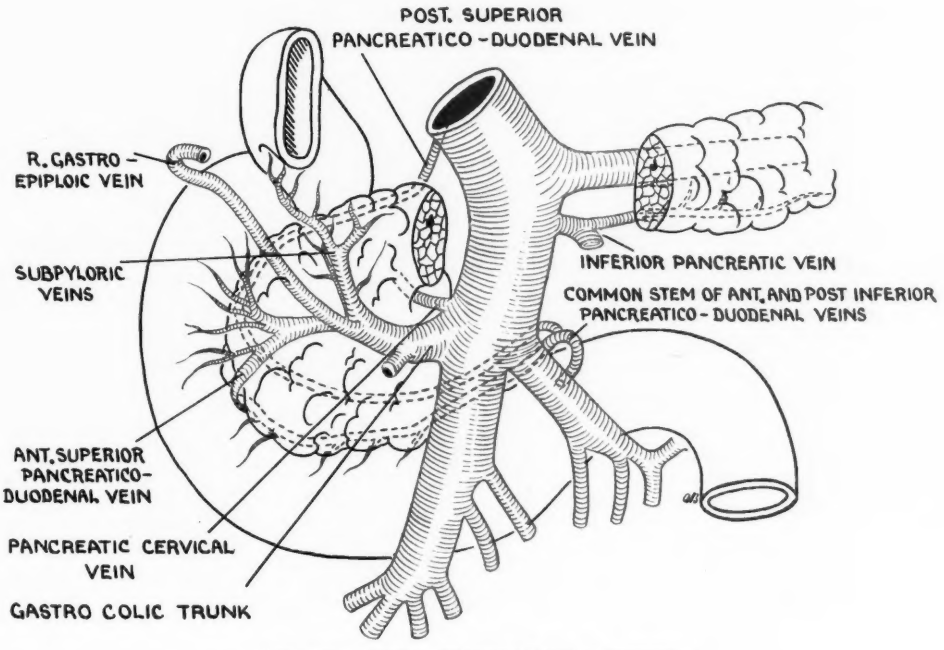

FIG. 447.—Venous drainage of head of pancreas.

its course it was joined by tributaries both from the gland and from the duodenal wall. These tributaries accompanied the finest arterial ramifications, and the vein itself coursed in front of its corresponding artery (*Fig.* 447).

It coursed medially, overlapped by the pancreas or buried in the gland substance, to the margin of the uncinate process, where it passed behind the superior mesenteric vessels to end in the left side of the first jejunal tributary, close to the junction of

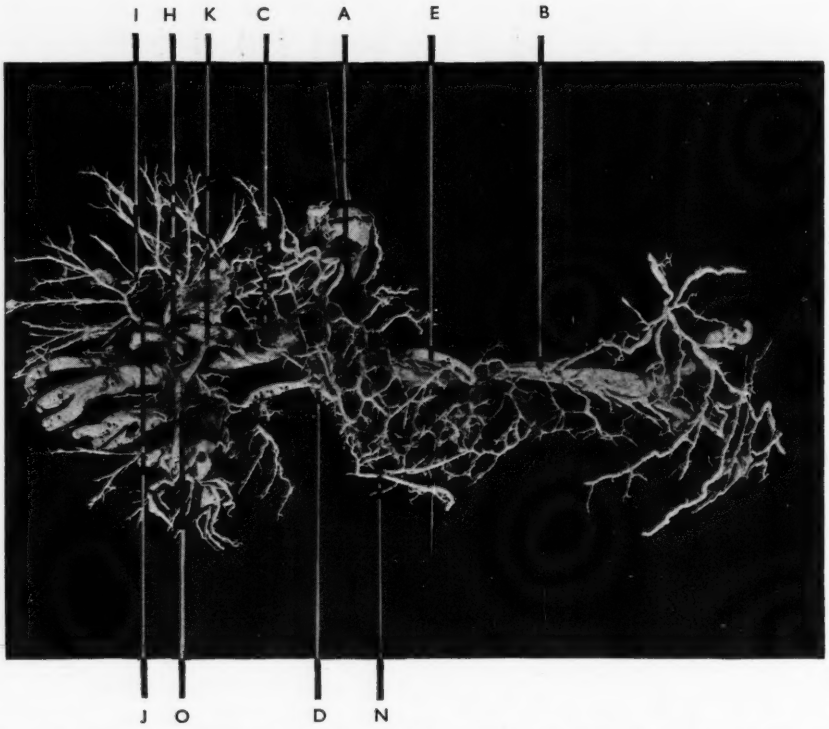

FIG. 448.—Anterior aspect of injection-corrosion preparation. (For key to vessels, *see Fig.* 445.)

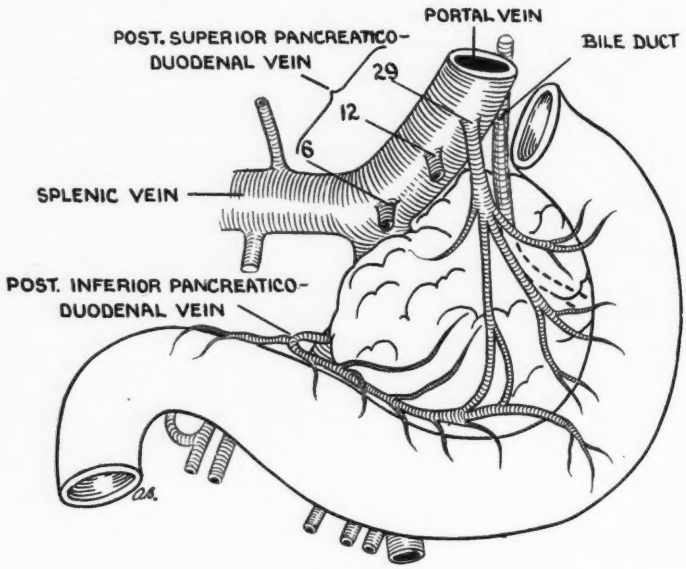

FIG. 449.—Posterior pancreatico-duodenal veins (posterior aspect).

The lower anterior part of the head and the adjoining duodenum were drained by the anterior inferior pancreatico-duodenal vein. This vein was formed in the sulcus near the origin of the superior vein, with which it usually established a venous arcade. that vein with the superior mesenteric trunk. This arrangement was present in 24 specimens; but in 11 instances the anterior inferior pancreatic vein joined the superior mesenteric trunk itself, entering its left side after passing behind the vein; in 9 instances

the posterior inferior pancreatic vein entered the first jejunal vein with it.

In an appreciable number of preparations the anterior inferior pancreatic vein was either absent far as the common bile-duct; thence it passed behind the bile-duct to join the posterolateral aspect of the portal vein. In 29 specimens its termination lay above the pancreas, in 12 at the level of the neck

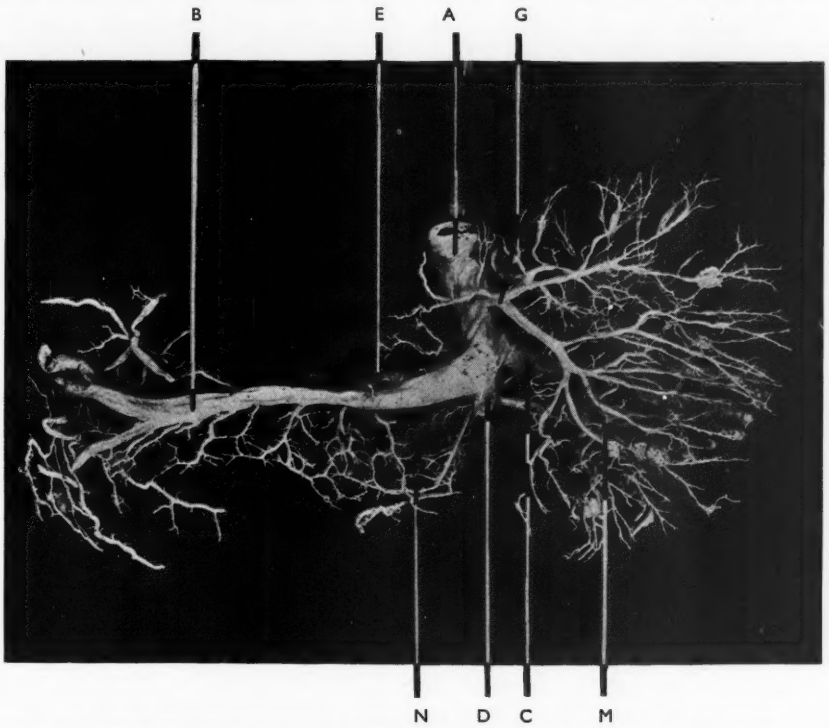

FIG. 450.—Posterior aspect of injection-corrosion preparation seen in *Fig.* 448. (For key to vessels, *see Fig.* 445.)

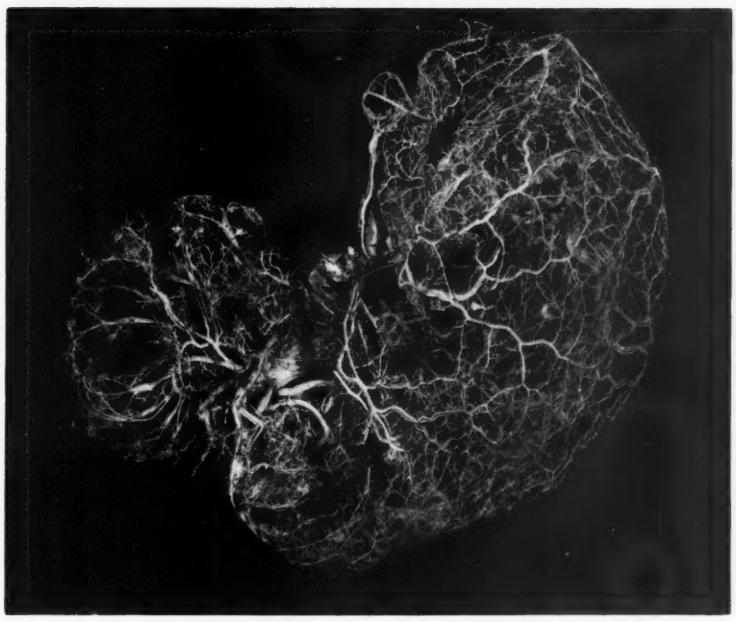

FIG. 451.—Complete injection of stomach, duodenum, and pancreas.

(12 cases) or insignificant, when the venous drainage was upwards to the superior vein.

The posterior aspect of the pancreatic head was drained by two main veins, which usually formed a well-marked arcade. The posterior superior pancreatico-duodenal vein accompanied its artery as of the pancreas, and in 6 immediately below the neck (*Figs.* 442, 446, 449). In 4 dissections the vein was duplicated; the upper vein ran superficial to its artery throughout its course, and passed between the duodenum and the common bile-duct to reach the portal vein, and the lower vein crossed

the posterior surface of the head to enter the portal vein behind the neck of the pancreas. (*Fig.* 442.) This arrangement was not noted in any of the injection-corrosion preparations.

The posterior inferior pancreatico-duodenal vein was present in 30 specimens. It originated in the duodenopancreatic sulcus below the common bile-duct, and drained blood from the lower part of the head of the gland and from the neighbouring part of the duodenum. Its course lay either in the sulcus or across the gland surface. It passed to the medial edge of the uncinate process around which it wound to reach its termination. In 19 of the injection-corrosion specimens it ended in the first jejunal tributary, sometimes with the anterior inferior pancreatico-duodenal vein, and in the dissected specimens also this was the most frequent finding. In 1 case the vein ended in the right limb joined a colic vein at its emergence from the mesocolon (*Figs.* 441, 447).

A pancreatic cervical vein, V. colli pancreatis (Kirk), was present in 9 specimens as a short wide trunk leaving the lower border of the pancreas, where the neck expanded into head and uncinate process. After a short course (0·5 cm.) outside the pancreas, it entered the posterolateral aspect of the superior mesenteric trunk, as it lay on the uncinate process, and before it passed behind the neck of the gland. This cervical vein was identified in 4 injection-corrosion preparations, and in 5 dissections (*Fig.* 447).

The remainder of the body of the pancreas was drained by a number (5 to 11) of small tributaries which entered the splenic vein directly.

The anterior middle pancreatic vein, described by Petren, was not seen in any of our specimens.

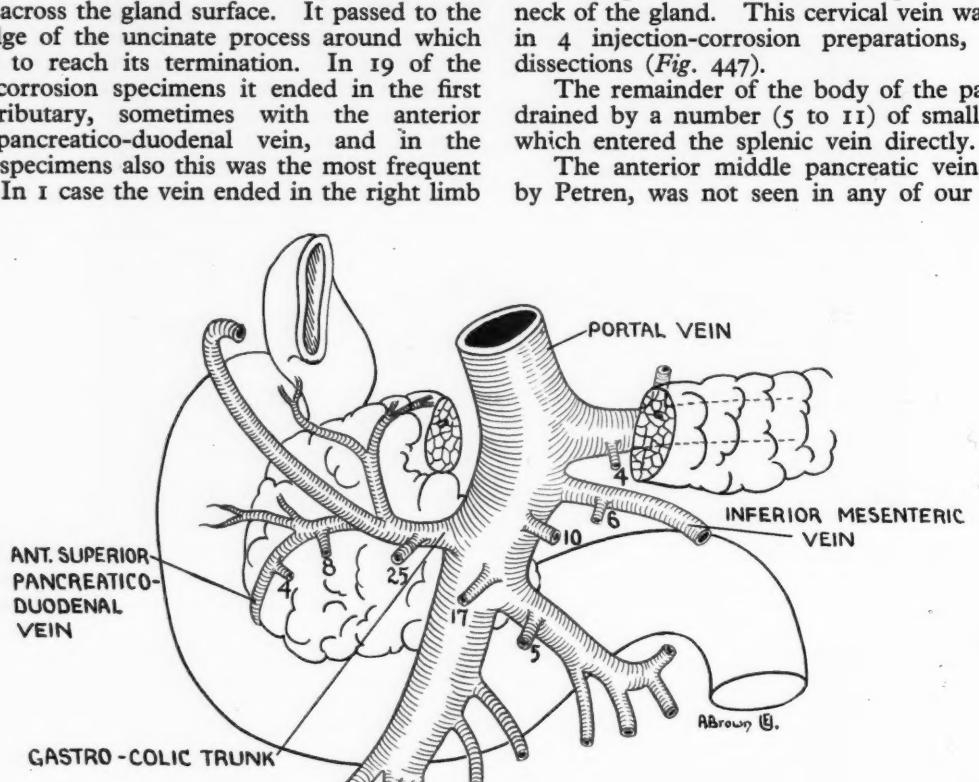

FIG. 452.—Frequency of termination of colic vessels.

of the superior mesenteric vein, and in 6 the main drainage of the lower part of the gland appeared to be upwards to the posterior superior pancreatico-duodenal vein (*Figs.* 446, 449).

In 5 dissected specimens it formed a common stem with the anterior inferior pancreatico-duodenal vein, so that this arrangement was encountered 9 times in the 30 specimens (*Figs.* 447, 451).

Venous Drainage of Neck and Body.—The inferior pancreatic vein was demonstrated in 31 specimens. A large vein collecting blood from the inferior part of the body of the pancreas, it came into view at the postero-inferior border of the body below the point at which the superior mesenteric vein passed under the neck to join the splenic vein. It had a variable termination, ending in the left border of the superior mesenteric vein in 13 specimens, in the inferior mesenteric vein in 11 specimens, in the splenic vein in 3 specimens, and in the gastrocolic trunk in 1 specimen. In the latter it adhered to the neck of the pancreas and crossed over the superior mesenteric vein. In 3 cases it

The Colic Veins.—The terminations of the colic veins in the region of the pancreas were noted, but their course in the mesocolon was not studied because the mesocolon had been removed at autopsy. The number of colic veins was variable, and on occasion as many as five could be recognized (*Figs.* 441, 452).

In the dissections, on occasion a distinction could be made between a middle colic and a right colic vein, but in others, and in the injection-corrosion specimens, it was not possible to differentiate between them. A constant termination for either the middle or the right colic vein could not be demonstrated.

For the most part the colic veins terminated in the gastrocolic trunk, and in the superior mesenteric vein; less frequently in the anterior superior pancreatico-duodenal vein, the splenic, or the inferior mesenteric veins.

From a study of all the preparations, it appeared that the veins entering the gastrocolic trunk and the anterior superior pancreatico-duodenal vein drained

the right half of the transverse colon and the right colic angle, whereas those ending in the superior mesenteric, splenic, or inferior mesenteric veins near their termination drained the middle and left portions of the colon, and appeared to accompany the middle colic artery.

In a few preparations a left colic vein was seen to pass medially below the pancreas to join the inferior mesenteric vein at right angles just before it ended in the splenic vein.

The Right Gastric Vein.—This vein was difficult to trace in the dissections, and its termination was identified in only 11 specimens. In 8 it entered the portal vein directly by passing between the hepatic artery and the common bile-duct; in 1 it passed obliquely upwards in the free margin of the lesser omentum, and entered the portal vein near the porta hepatis; and in the remaining 3 specimens the vein was traced as far as the porta hepatis without demonstrable union with the portal vein (*Figs.* 441, 442).

In corrosion preparations, tributaries from the pylorus and duodenum could be traced to the posterior superior pancreatico-duodenal vein, which they joined immediately before it entered the portal vein. The pyloric vein of Mayo was found to drain into this vessel in several preparations.

Summary.—

1. The left gastric vein ended in the portal vein (20 cases), in the splenic vein (15 cases), or in the splenoportal junction (4 cases).

2. The inferior mesenteric vein ended in the splenic vein in 26 cases, and in the superior mesenteric vein in 17 cases.

3. The right gastro-epiploic vein united with the anterior superior pancreatico-duodenal vein and, later, with a colic vein to form a gastrocolic trunk, which terminated in the superior mesenteric vein.

4. Anterior and posterior venous arcades are invariable features of the vascular arrangements around the pancreas.

5. An inferior pancreatic vein was present in 31 cases.

6. A pancreatic cervical vein was an occasional finding (9 cases).

7. The colic veins were frequently duplicated and were variable in their terminations.

8. The right gastric vein was small and ended in the portal vein, in the posterior superior pancreatico-duodenal vein, or by passing upwards in the free edge of the lesser omentum to the porta hepatis.

DISCUSSION

In the standard text-books of anatomy descriptions of the vascular arrangements of the pancreas, and especially of the venous drainage, are not particularly helpful to the surgeon called upon to undertake radical surgery in this difficult region.

In this study we have attempted a more detailed description and sought to determine the surgical importance of the several vessels.

It is a striking fact that, despite the close relationship of the neck of the pancreas to the portal vein, in no case did a pancreatic tributary enter the portal vein on its anterior aspect. It follows that separation of the neck in this plane during the operation of pancreatico-duodenectomy is a procedure without hazard, unless the pathological process for which the operation is undertaken has involved the neck and extended to become adherent to the vein itself. The majority of carcinomata of the pancreas arise to the right of this area, and in these cases the division of the neck could be accomplished without difficulty.

In the usual operation for pancreatico-duodenectomy the duodenum is transected in its third part, the fourth part and the duodenojejunal flexure being left in situ. Our studies have shown that in cases in which the uncinate process was large, it extended well behind the mesenteric vein and lay in close apposition to the superior mesenteric artery. In such cases the common trunk of the inferior pancreatico-duodenal and jejunal arteries was very short, and during the mobilization of the head of the gland it might be necessary to ligate the vessel proximal to the jejunal branch, with a considerable risk of gangrene of the flexure, a fact which has been noted by other workers (Pierson, 1943; Ziegler, 1943). In such cases it would be necessary to modify the operation by removing the fourth part of the duodenum and the flexure. In certain clinics this step is now an established part of the radical operation.

The occasional presence of a large hepatic branch of the superior mesenteric artery in relation to the posterior aspect of the head must be borne in mind when mobilizing the head of the gland. Here the vessel is particularly liable to be involved in disease of the pancreatic head, and in this event the head could not be freed without injury to the vessel and considerable hæmorrhage. The fact that the vessel may be the only hepatic artery has an additional significance.

Easy and safe mobilization of the head might also be difficult on account of the pancreatic cervical vein, which invariably entered the right side of the superior mesenteric vein just below the neck and usually slightly posteriorly, and was thus hidden from the front.

Our observations seem to indicate that it would be possible to control the main efflux from the anterior aspect of the head by ligation of the gastrocolic trunk formed by the confluence of the right gastro-epiploic, colic, and anterior superior pancreatico-duodenal veins. The pancreatic tributaries of the splenic vein have virtually no extraglandular course; in consequence, ligation of these would be difficult and, in the presence of a pathological condition in this part, impossible. This consideration is not important in the usual operation for cancer of the head of the pancreas, but it is important in complete removal of the pancreas, when total pancreatectomy is required, and in some clinics it is now customary to remove the spleen with the pancreas.

The other points which emerged from this study are more particularly of anatomical interest. The frequency and size of the inferior pancreatic vein would seem to justify reference to it in text-books of anatomy. The variable termination of the left gastric and inferior mesenteric veins is also worthy of note; the former ended in the splenic vein or the splenoportal junction almost as often as

the portal vein itself, while the inferior mesenteric vein in many cases joined the superior mesenteric vein rather than the splenic.

The occasional formation of the superior mesenteric vein by the union of the right and left trunks in front of the artery immediately below the splenic vein does not appear to have been previously reported, and would seem to have some importance in view of the occasional use made of the superior mesenteric vein in the various forms of shunt for portal hypertension.

GENERAL SUMMARY

A study of the blood-vessels of the pancreas in 27 dissections and in 23 injection-corrosion preparations has been undertaken.

The arterial arrangements were found to differ but little from the descriptions available in the literature.

The large venous trunks were found to be constant; the smaller trunks varied considerably, but within recognizable limits.

A plea has been made for the inclusion of some of the more constant findings in works on surgical anatomy.

We thank Professor Sir James Learmonth, Mr. John Bruce, and Professor Ian Aird for their stimulus and advice during this investigation. The dissections were carried out by one of us (C. W. A. F.) under the direction of Sir James Learmonth and Mr. Bruce in the Department of Surgery of the University of Edinburgh, and the injection-corrosion preparations were made by the other (E. G.) under the direction of Professor Aird in the Department of Surgery, Post-graduate Medical School, London.

REFERENCES

BRUN, H. (1913), *Beitr. klin. Chir.*, **84**, 305.
DESCOMPS, P., and LALAUBIE, G. de (1912), *J. Anat., Paris*, **48**, 337.
EDWARDS, L. F. (1941), *Anat. Rec.*, **81**, 351.
FLINT, E. R. (1922), *Brit. J. Surg.*, **10**, 509.
HALLER, A. (1748), *Disputationum Anatomicarum Selectarum.* Gottingen: Vandenhoeck.
KIRK, E. (1932), *Z. ges. Anat.*, **94**, 822.
MORISON, D. M. (1923), *J. Amer. med. Ass.*, **81**, 174.
PETREN, T. (1929), *Z. ges. Anat.*, **90**, 234.
PIERSON, J. M. (1943), *Surg., Gynec. Obstet.*, **77**, 426.
TONKOFF, V. N. (1903), *Russkij. Wratch*, **20** (quoted by KIRK).
TRUETA, J., BARCLAY, A. E., DANIEL, P. M., FRANKLIN, K. J., and PRICHARD, M. M. L. (1947), *Studies of the Renal Circulation.* Oxford: Blackwell.
VERNEUIL, A. (1851), *Gaz. Méd. Paris*, **6**, 384.
WHARTON, G. K. (1932), *Anat. Rec.*, **53**, 55.
WIART, J. (1899), *J. Anat., Paris*, **1**, 104.
WILKIE, D. P. D. (1911), *Surg., Gynec. Obstet.*, **30**, 374.
YULE, E. (1927), *J. Anat., Lond.*, **61**, 345.
ZEIGLER, H. P. (1943), *Surg., Gynec. Obstet.*, **74**, 137.

NASOPHARYNGEAL TUMOURS

THE PLACE OF LEUCOTOMY IN THE TERMINAL STAGES OF THE DISEASE

BY E. C. MEKIE AND GORDON RANSOME, SINGAPORE

MALIGNANT tumours of the nasopharynx are reported from all parts of the world, but their incidence amongst members of the Chinese race is exceptionally high. In Singapore, with its predominantly Chinese population, this tumour ranks third in frequency amongst neoplasms.

While without full pathological conviction, we are in the habit of employing the name coined by Regaud (1921) and by Schminke (1921) of 'lymphoepithelioma'. The description which they gave of the essential pathological features corresponds to the histological characteristics of the lesion so commonly encountered in this country.

Macroscopically the tumour is often a bulky lesion which appears to originate in the posterior wall of the nasopharynx, filling the whole of that space and then extending into the nose and pterygoid fossæ. Intracranial metastases are common. The extension into the cranium presents many points of interest. Not infrequently large intracranial deposits are present without radiological evidence of skull erosion, while in post-mortem specimens direct connexion between the pharyngeal tumour and the intracranial deposits is not always obvious. Bony destruction is frequently not marked. Gardham (1929) and others have stressed the manner in which this tumour can extend into the cranial cavity via the foramina of the base of the skull, especially by the jugular foramen, the carotid canal, and the foramen spinosum. Recently we have had the opportunity of studying two specimens in which there was clear evidence of transgression of the occipito-atlantoid membrane. Within the skull the

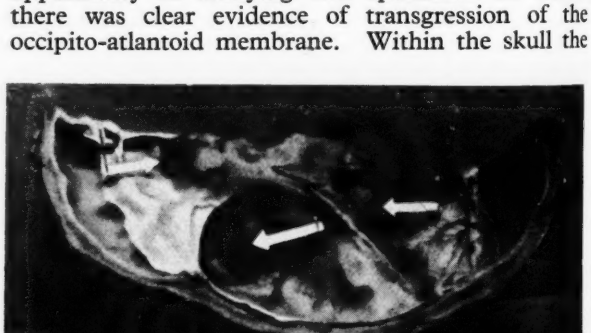

FIG. 453.—Post-mortem specimen from a case of lymphoepithelioma demonstrating the extradural deposits in the middle and posterior fossæ.

deposits are found to be extradural in location (*Fig.* 453). This point was stressed by Reverchon in 1923. We have not seen examples of secondary tumours occurring within the brain tissue itself.

The later stages of this terrible disease are distressing in the extreme. In addition to the foul

discharge and hæmorrhage from the primary tumour, there is difficulty in eating, breathing, and phonation. The secondary deposits in the skull produce pressure on the cranial nerves, with resulting palsies and neuralgias. Due probably to the stretching of the dura over the tumour masses, there is intense headache, usually localized in the frontal and parietal regions. The continuous intense pain, preventing

FIG. 454.—*Case* 2. Case of lympho-epithelioma of nasopharynx. Appearance of patient on admission.

rest or sleep, rapidly undermines the morale, and the general condition of the patient deteriorates. They sit crouching in corners, holding their heads in their hands, dejected and miserable until death releases them. But death may be delayed, for in only a minority of cases is the progress of the tumour sufficient to prove fatal in a short period of weeks. We have not found any drug which is potent enough or retains its potency to make the pain bearable.

While these tumours are radiosensitive, recurrence has been, in our experience, very common and rapid. The work of Freeman and Watts (1946) and Scarff (1948) suggested that benefit might follow prefrontal leucotomy. These workers had been able to secure marked amelioration of pain in cases of recurrent carcinoma and diseases of the central nervous system, but it was not clear whether relief would be obtained when the lesion causing the pain was within the cranium and largely, we believed, due to meningeal tension. Another consideration which made the operation justifiable was the fact that we were of opinion that truly intracerebral metastases were most uncommon.

Four cases of advanced disease have now been submitted to bilateral prefrontal leucotomy. Brief case notes are appended.

CASE REPORTS

Case 1.—Male Chinese, aged 34. Eight months' history of numbness of the right side of the face and gradually increasing blindness in the right eye which had proceeded to total loss of vision. Difficulty in swallowing for three months. There was ptosis on the right side and the tongue deviated to the right. There was intense pain in the right eye and right frontal area.

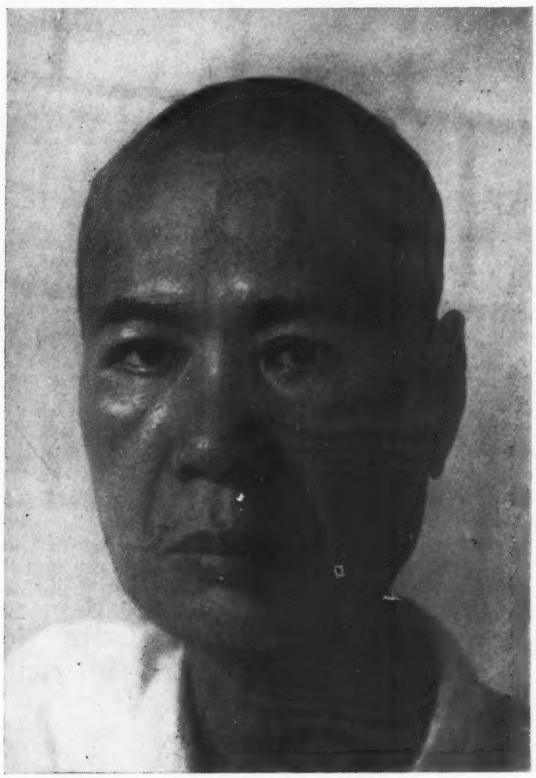

FIG. 455.—Same case. Appearance of same patient ten days after prefrontal leucotomy.

There was an enlarged cervical node on the left side at the level of the angle of the jaw. Biopsy report: Cervical gland showing infiltration with lympho-epitheliomatous tumour.

Prefrontal leucotomy performed. Patient died four hours after operation. At post-mortem flat masses of new growth were found in the middle and posterior fossæ on the right side. The primary tumour in the nasopharynx was found to extend into the palate and base of the skull. The exact immediate cause of death was not ascertained.

Case 2.—Male Chinese, aged 53. Swelling below the angle of the jaw for three months. Severe headache in the left parietal region with facial neuralgia. There was inability to open the mouth. Examination showed a large ulcerating tumour of the nasopharynx pushing down the palate on the left side and invading the fauces. An enlarged and hard lymph-node was present on the left side, lying posterior to the angle of the jaw and fixed to the surrounding tissues.

The general condition of the patient was miserable. He sat on his bed with bowed head and never spoke to anybody. Sleep was only secured with difficulty by the use of full doses of opiates.

Prefrontal leucotomy performed. For the first few days it was difficult to ascertain the effect of the operation, but from the fourth day onward he improved rapidly. He asserted that the pain was still present but did

so with a smile and he became more 'normal' in his behaviour and relations with other patients. His general condition improved rapidly (*Figs.* 454, 455).

Case 3.—Male Chinese, aged 42. Swelling appeared behind the jaw on the right side of the neck, four months before admission. This was associated with severe pain in the head and on the right side of the face. A second swelling appeared some weeks later on the left side of the neck. Internal strabismus of the right eye was noted a month before admission. There was inability to open the mouth and the examination of the nasopharynx was impossible.

The general condition was poor. Like the previous case he was morose and silent.

Biopsy from a node in the neck revealed "lympho-epithelioma". Following prefrontal leucotomy he became decidedly better. He began to converse with the other patients and, while still asserting that he had pain, he did not complain about it unless asked. He no longer sat about holding his head.

Case 4.—Male Chinese, aged 60. The history in this case was of pain over the vault of the head for a period of three months. This had become progressively worse and he could not sleep at night. During the same period there had been a progressively increasing swelling in the upper part of the left side of the neck, which on examination was a group of typically hard adherent malignant lymph-nodes.

There was inability to open the mouth fully and examination of the nasopharynx was limited, but a large tumour could be felt behind and above the palate. The general condition was poor. After prefrontal leucotomy there was a dramatic disappearance of the headache within twenty-four hours. He asserted, in spite of suggestion, that the old headache had completely disappeared, but he complained occasionally of pain at the site of the incisions.

GENERAL APPRECIATION OF THE RESULTS OF OPERATION

(G. R.)

The above brief description of the disease gives but a poor idea of the suffering experienced by these patients. In time past one's therapeutic armamentarium has been taxed to the full in order to try to provide some relief, usually without success, as many of the patients have previously been opium addicts. The prolonged suffering undermines not only the morale of the sufferer but to a certain extent that of the doctor and the ward staff as it gives rise to such a feeling of helplessness in the face of disease. Leucotomy, in the three cases which were successful, has provided, in our opinion, the most valuable method of amelioration we have yet encountered. Two of the patients became relieved of their pain almost at once, and very soon began to walk about the ward and chat to other patients. They began to eat their meals with relish, something they had not done for weeks, with resulting increase in weight. They even were able to smile and laugh, and in a few days one could scarcely recognize them for the miserable objects they had been previously. While relieved of their pain and discomfort, none of the three successful cases has demonstrated a mental deterioration of any marked degree, nor one which would occasion any distress to relatives.

To prolong life and prevent suffering are the twin functions of the physician, and while in the later stages of this form of malignant disease the first may be beyond us, we believe that in leucotomy we have a measure which enables us to do much to attain the latter.

Acknowledgement is made of the permission of the D.M.S., Singapore, to report these cases.

REFERENCES

FREEMAN, W., and WATTS, J. W. (1946), *Lancet*, **1**, 953.
GARDHAM, A. J. (1929), *Brit. J. Surg.*, **17**, 242.
REGAUD, M. (1921), *Bull. Soc. franç. Oto-rhino-lar.*, **34**, 209.
SCARFF, J. E. (1948), *J. Neurosurg.*, **5**, 288.
SCHMINKE, A. (1921), *Zieglers Beitr.*, **68**, 161.

LYMPHANGIOMA OF THE KIDNEY

BY D. WYNN-WILLIAMS AND A. D. MORGAN

A SEARCH through the literature having failed to reveal a similar clinico-pathological picture, the following case seems worthy of record.

CASE REPORT

HISTORY.—G. B., a female, aged 33, and married for seven years at the time when first seen, was found to be 23 weeks pregnant with the first baby. She was attending a large maternity clinic in London, and after passing a specimen of urine for her routine monthly visit noticed it to be red and to contain clots of blood. She was uncertain whether this originated from the bladder or vagina. Owing to pressure of work she failed to attend hospital that day, and as the urine was normal in the evening she postponed her visit. Seven days later she again noticed red discoloration of her urine and this time reported to the hospital. She was admitted as an emergency the same day with a possible diagnosis of ante-partum hæmorrhage; but a catheter specimen of urine that evening showed hæmaturia with many red cells. In view of these findings she was transferred to Westminster Hospital for urinary investigation. During the week between the first hæmaturia and reporting to hospital, she complained of severe cramp-like pains generalized over the abdomen that were unrelated to micturition and passed off when resting.

Personal History.—Micturition: Frequency since pregnancy, $\frac{D}{N} = \frac{3-4}{1}$

Had noticed one year before a red-coloured urine on one day, associated with generalized abdominal pain, but as it cleared up spontaneously had thought no more about it.

Bowels: Constipated and took daily laxatives.

Menses: L.M.P. March 7, 1947.

Past Illnesses.—Rheumatic fever 13 years of age; scarlet fever and diphtheria as a child; appendicectomy at 19 years of age for recurrent appendicular pain.

ON EXAMINATION (Sept. 6).—A stout female—weight 11 st. Not clinically anæmic. Respiratory, cardiac, central nervous systems N.A.D. B.P. 125/75. *Abdomen*: Enlarged uterus palpable and fœtus presentation R.O.A.,

and a tender mass palpable in R.I.F.—probably a degenerating fibroid.

INVESTIGATIONS.—

Sept. 9: *Cystoscopy*—Basal cystitis. Efflux of blood from right ureteric orifice, with distortion of right side of bladder as from a mass pressing from outside.

Sept. 16: *Retrograde pyelography—right.* A small quantity of contrast medium has entered the right renal pelvis, outlining a tumour which is distorting the cavity. There is considerable dilatation and distension of the pelvis and calices (*Fig.* 456).

Right ureteric specimen of urine: Blood cells and a few pus cells. No organisms. No growth on culture. Wassermann and Kahn reactions, negative.

Sept. 17: *Intravenous pyelography*—Single fœtus—transverse lie.

Left kidney—duplex pelvis and double ureter—secreting well.

Right kidney—no excretion.

It was decided on these findings that the provisional diagnosis was that of a hypernephroma or a large papilloma of the pelvis in the right kidney and an exploration was, therefore, decided upon.

AT OPERATION (Sept. 20).—*Right nephrectomy.* Under pentothal, gas, oxygen, and cyclopropane the patient was placed in the left lateral position. Considerable difficulty was experienced in positioning the patient, as it was inadvisable to break the table excessively or allow pressure from an abdominal pad in view of the pregnancy.

FIG. 456.—Right retrograde pyelography. Intrapelvic tumour outlined. Considerable dilatation of pelvis and calices.

A right renal incision was made, the musculature incised, and the kidney exposed. The surface of the kidney was found to be normal in consistency and the capsule intact. Great difficulty was experienced in freeing the kidney owing to the proximity of the 12th rib and the crest of the ileum. This necessitated the resection of the 12th rib.

On manual examination of the kidney a tumour was palpated in the pelvis, confirming the radiological appearances, and the kidney was removed together with 2 in. of the ureter.

The wound was closed in layers and a drainage tube inserted.

PROGRESS.—

Post-operatively.—Convalescence was complicated by distension and right-sided abdominal pain of a severe nature, requiring opiates and enemata. The wound healed well following removal of the drainage tube and the patient was finally discharged on the twenty-third post-operative day. The fœtus was alive and the fœtal heart-sounds normal.

Follow-up.—On discharge the urine was found to be normal. Intravenous pyelogram two months after operation—left kidney functioning normally. Blood-urea 61 mg. per cent. Plasma-chlorides 580 mg. per cent. Alkaline reserves 49 c.c. per cent. Liver function tests—all normal.

FIG. 457.—Vertical section of kidney. Honeycomb appearance with many small cysts. (*No magnification.*)

Pregnancy continued uninterrupted and the patient was delivered of a normal female child in December, 1947. Following the delivery, the patient developed a continuous dull right-sided abdominal pain which was present following discharge from hospital, but not severe enough for her to report to her doctor. In February, 1948, the pain became acute and generalized over the whole of her lower abdomen. A diagnosis of acute abdomen was made and she was again admitted to Westminster Hospital. Laparotomy was performed under a general anæsthetic and revealed a right twisted ovarian cyst, which was removed. Her recovery was uninterrupted and she was completely relieved from the previous abdominal pain.

PATHOLOGY.—

Naked-eye Appearances.—The kidney as a whole was smaller than normal, and only 8·5 cm. from pole to pole, though the renal pelvis was obviously distended by a tumour, two-thirds of which lay within the renal part of the pelvis, around which the pale and atrophic remains of the kidney substance appeared to form a capsule, devoid of pyramids or calices; the rest lay within the pelvic portion, blocking the outlet. The tumour was in the form of a polypoid growth, originating in the renal part of the pelvis, to which it was attached near the upper pole by a short pedicle 2 cm. broad; otherwise it was quite separate from the pelvic lining, though its smooth white surface fitted neatly into the pelvic cavity, which clearly had moulded the tumour during its growth.

When cut across (*Fig.* 457), the tumour presented a honeycombed surface consisting for the most part of small cysts ranging from 1 to 8 mm. in diameter, transparently thin-walled except for those near the centre, which were embedded in a denser opalescent stroma. All contained clear fluid, and some of the larger cysts were loculated.

Microscopically (*Figs.* 458–460).—The cysts were lined by a single layer of flattened cells resembling endothelium. In some of the spaces the albuminous residue of their fluid contents remained in paraffin sections, but in none of them was blood present. The intervening septa contained strands of collagen with occasional smooth muscle-fibres and thin elastic fibrils, these elements being grouped around the smaller cysts in a concentric fashion. The supporting blood-vessels were small and thin walled. A puzzling feature was the presence of occasional groups of acinar structures in the stroma, apparently lined by cubical epithelium. Their occasional elongated appearance suggested that they might be surviving collecting tubules, and it is worthy of note that these structures were most abundant in the fibrous pedicle between the cysts and the surviving kidney tissue. The latter consisted of a narrow zone showing gross atrophy with fibrosis, but throughout which a few glomeruli were clearly recognizable. These showed thickening of the capsular basement membrane and increased patency of the glomerular capillaries, such as is often seen in long-standing hydronephrosis. Scanty hyalinized forms were observed. The renal tubules had undergone almost total atrophy, and it was impossible to define the cortico-medullary junction. Signs of inflammation were, in general, absent, but occasional groups of lymphocytes were present. The medium-size arteries and arterioles showed no appreciable degree of thickening of their walls.

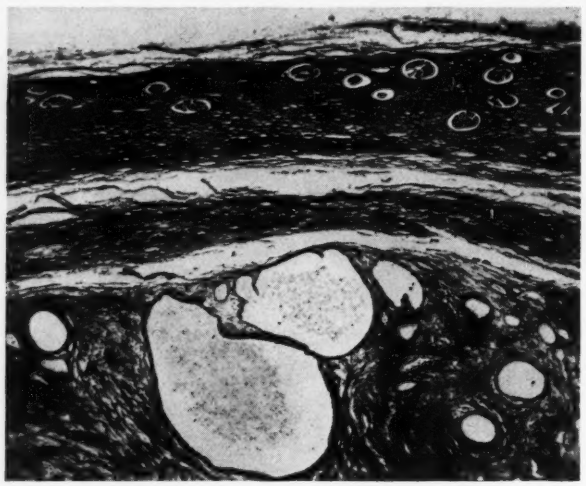

FIG. 458.—Low-power view of kidney showing atrophic renal cortex stretched around the tumour. Hæmatoxylin and van Gieson. (× 30.)

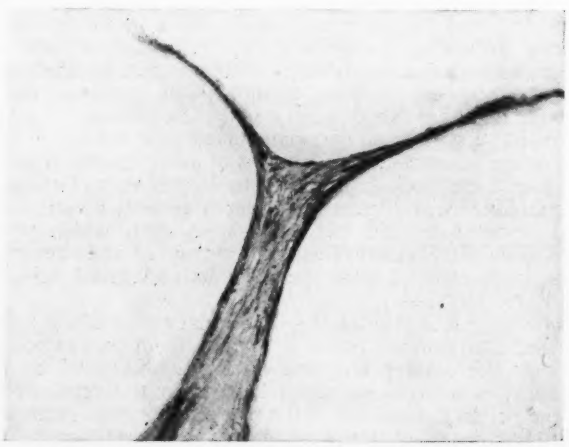

FIG. 460.—Elastic tissue in the walls of three contiguous cysts Weigert's elastic stain. (× 100.)

DISCUSSION

Lymphangioma of the kidney has been recorded so infrequently in medical literature that the diagnosis is advanced with some diffidence. Polycystic disease, and what has been called ' cystadenoma ', have been considered and discarded in view of the musculo-elastic tissue in the cyst walls and the fact that a study of recorded cases of these conditions yielded a picture quite unlike the present one.

FIG. 459.—In the interstitial tissue of the tumour, between small cysts, are structures resembling renal tubules. Hæmatoxylin and eosin. (× 52.)

In 1866 Henschel described a renal lymphangioma in a female aged 63, but though it was " the size of an apple " it did not contain definite cysts. Perlmann in 1928 gave an account of a polycystic tumour of the left kidney in a female aged 55, causing downward displacement of the kidney and compression of the renal pelvis as seen in the pyelogram. When removed, it was found to be globular in shape, 11 cm. in diameter, and bulging from the convex border of the kidney, the upper pole of which was stretched around the tumour like a capsule. Medially it was compressing the renal pelvis. Microscopically the cysts were lined by flattened epithelium, and hæmorrhage had occurred into some of the spaces. The author, in diagnosing lymphangioma, pointed out that as a group these tumours contain both elastic and muscle fibres in the interstitia.

Willis in 1948 described a lymphangioma of the kidney, in a female aged 67, replacing a small part of the centre of the organ and projecting laterally from it. On section it showed a honeycomb of large and small cysts separated by fibrous septa and containing watery fluid. Microscopically the cysts were lined by flat non-epithelial cells.

A number of cystic *hæmangiomata* of the kidney have been described (e.g., Jacobs and Rosenberg, 1927; Jenkins and Drennan, 1928; Gile, 1929; Judd and Simon, 1928), and it is interesting to note that in some of these a number of empty cysts were

observed, which raises the possibility that a proportion of these tumours may in fact have been lymphangiomata, since hæmorrhage into a lymphangioma is frequently a means of directing attention to an unsuspected tumour.

From a clinical aspect the diagnosis of hæmaturia of sudden onset is of considerable interest, and this case demonstrates the necessity for full and immediate investigation. The urgency is mainly from the possibility of a malignant neoplasm being associated with pregnancy. The diagnosis of renal tumour, either hypernephroma or papilloma of the pelvis, is the only one which could have been made preoperatively. The causation of the hæmaturia itself must, in view of the pathology, be a matter of conjecture, and is probably due to the distension or rupture of some small veins over the surface of the tumour ; or even rupture of one of the cystic spaces following some unsuspected minor trauma. The operation of nephrectomy during the middle three months of pregnancy is in accordance with the statements made that this period is probably the safest in which a major operation should be carried out. The acute abdominal pain suffered post-operatively and following delivery of the child was in all probability due to the ovarian cyst and entirely unrelated to the lymphangioma. The fact that the cyst was undiagnosed throughout pregnancy and indeed until twisting occurred, made post-operative care extremely difficult.

SUMMARY

1. A case of hæmaturia in a pregnant woman is presented.

2. The difficulty of diagnosis is discussed, the only possibilities being that of a renal tumour involving the pelvis, probably hypernephroma or papilloma of the pelvis.

3. The final diagnosis is only made possible on exploration of the kidney.

4. A polypoid cystic tumour distending and obstructing the renal pelvis and causing fibrous atrophy of the kidney is described.

5. Pathologically the tumour presented the characters of a lymphangioma.

The clinico-pathological picture appears to be unique in the literature.

We are indebted to Mr. G. T. Mullally for permission to publish the case, and to the Westminster Hospital Photographic Unit for photographs and reproduction of the radiograph.

REFERENCES

GILE, H. H. (1929), *Surg., Gynec. Obstet.*, **48**, 555.

JACOBS, P. A., and ROSENBERG, W. (1927), *J. Urol.*, **17**, 337.

JENKINS, J. A., and DRENNAN, A. M. (1928), *Ibid.*, **20**, 97.

JUDD, E. S., and SIMON, H. E. (1928), *Surg., Gynec. Obstet.*, **46**, 711.

A CASE OF COLON AND ILEUM DUPLEX

BY JOHN AITKEN, GLASGOW

NUMEROUS cases of duplication of bowel have been described in the literature, but it is felt that the present case, which shows many unusual features, should be recorded.

CASE REPORT

M. F., female, 2 hours old, was admitted on March 12, 1949, to the Royal Hospital for Sick Children, Glasgow, as an emergency.

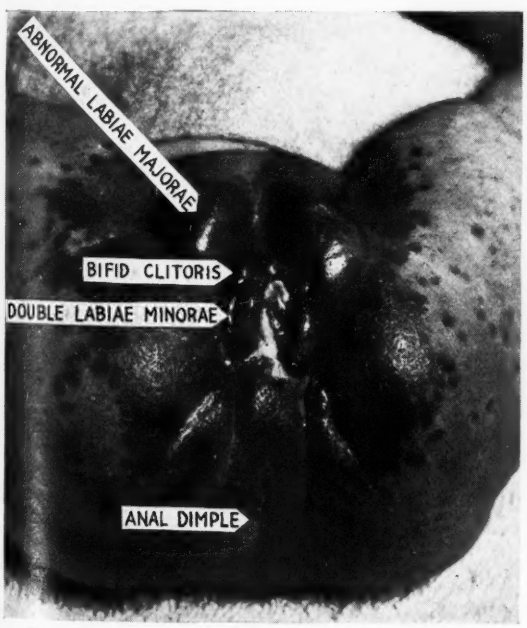

FIG. 461.—This shows the various abnormalities of the perineum.

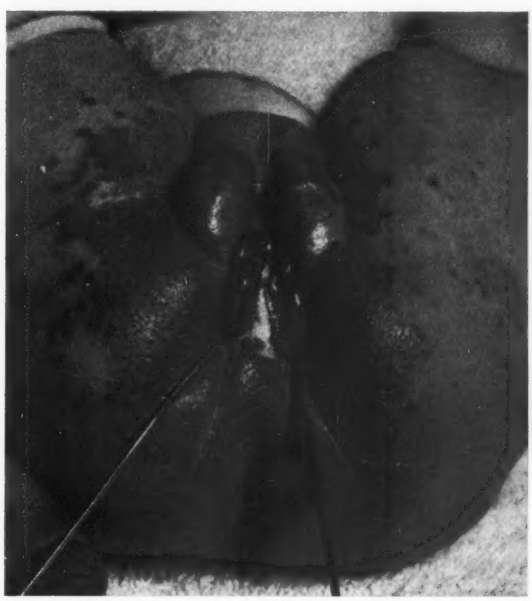

FIG. 462.—The perineum. Probes have been inserted into the rectal fistulæ.

ON EXAMINATION (*Figs.* 461–463).— The infant, 8½ lb. in weight and apparently healthy, presented the following features :—

1. A large exomphalos, about the size of half a grapefruit, through the sac of which coils of bowel could be seen.

2. Perineum—labia majora present in their anterior halves only. Two pairs of deformed labia minora lay

FIG. 463.—The perineum. Probes have been inserted into the vaginal openings.

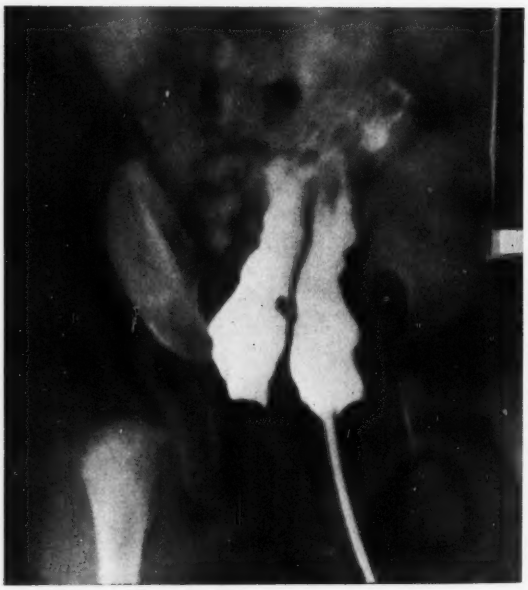

FIG. 464.—Lipiodol. This shows double rectum.

posteriorly and medially to these. The clitoris lay in two halves and the urethra opened at the root of the bifid organ.

Two openings were present between each pair of labia minora. These were vaginæ anteriorly and recto-vaginal cloacæ posteriorly. Meconium issued from both recto-vaginal cloacæ. There was no anus ; only a furrow scarcely amounting to an anal dimple was present. There were no obvious signs of the presence of a sphincter.

FIRST OPERATION.—The exomphalos was operated upon the same day in the standard manner. It contained small and large bowel only. No difficulty was experienced in replacing these into the abdomen until the final loop, which was found to be adherent to the apex of the exomphalos sac and had to be freed by sharp dissection, the vitelline artery being divided and ligated in the process. When this had been completed, the adherent area was found to be a sac of bowel about 1 × 1½ in. in size, from which ran three loops of small intestine. The infant's condition did not allow of further investigation.

A presumption was made that the bowel was duplicated from the sac (i.e., the apex of the midgut loop) as far as the two openings in the perineum.

FURTHER INVESTIGATIONS (*Figs.* 464–466).—

1. Double lipiodol enema.
2. Double barium enema.

} These showed the rectum and colon to be duplicated as far as the hepatic flecture.

3. Barium meal : The abnormalities of the gastro-intestinal tract were not shown.

Experience in this hospital in cases with bowel duplication and in cases of rectovaginal cloaca has shown that the duplications are prone to enteritis and hæmorrhage, usually with a fatal outcome, and that the cases of rectovaginal cloaca develop serious complications as a result of fæcal impaction. In these latter cases we have found it advisable to perform colostomy within a few weeks of birth, otherwise the fæcal impaction gives rise to gross hypertrophy and lengthening of the large bowel.

The following procedures, therefore, were contemplated in this case :—

1. Immediate resection of whichever duplication was feasible from apex of mid-gut loop to pelvic floor.
2. Colostomy one to two weeks later.

FIG. 465.—Barium enema. P.A. view. Double colon visualized as far as hepatic flexure.

3. At the age of 4–5 years transplantation of cloaca posteriorly into true anal site through the sphincter if present.

A laparotomy with the first of these ends in view was performed on April 25.

SECOND OPERATION.—The findings were as follows (*Figs.* 467, 468) :—

1. Colon duplex, pelvic floor to cæcum.

2. Duplication of cæcum.
3. Duplication of appendix.

All these organs had a common muscular and peritoneal coat.

4. A sac of bowel the size of a ping-pong ball was present at the apex of the mid-gut loop, over which ran the remnant of the vitelline artery, this corresponding with the site of the vitello-intestinal duct and presumably being a type of Meckel's diverticulum (*Fig.* 468).
5. Normal ileum entered this sac at its proximal end.
6. Two loops of ileum left the sac at its distal end. These loops were of equal diameter and length, each having a distinct mesentery and each ending by entering one half of the double cæcum.
7. The uterus was felt to have an anteroposterior groove across its dome and was presumably of the bicornate type. Right and left ovaries and tubes appeared to be normal.

The plan of resecting one half of the duplicated bowel had to be abandoned owing to the colon and cæcum having common muscular and peritoneal coats, and the abdomen was closed without further interference.

FIG. 466.—Barium enema. Lateral view.

DISCUSSION

I cannot find any case in the literature quite like this one. Duplication of the bowel commencing in a Meckel's diverticulum has not, as far as I can find, been described before, and must be extremely rare.

The prognosis in this case is, of course, extremely bad, and an early fatal outcome is likely from various causes, such as enteritis, hæmorrhage from ulceration of the diverticulum, volvulus, or intestinal obstruction.

It is doubtful whether any surgical procedure would result in a normal functioning bowel and anus quite apart from the question of the double vagina and abnormal genital tract. Should the child survive to a reasonable age resection of the entire gut from proximal to the diverticulum to proximal to the pelvirectal junction, with anastomosis and thereafter a plastic operation on the cloaca, will have to be considered.

SUMMARY

A case of duplication of the bowel from the apex of the midgut loop to the perineum is described, the duplication starting in what appears to be a rare type of Meckel's diverticulum. This is associated with double rectovaginal cloacæ, double vaginæ, and a bicornate uterus.

I wish to thank Mr. A. P. Laird, in whose wards the infant was treated, for his encouragement and permission to publish the case; and Mr. C. Palmar for the photographs.

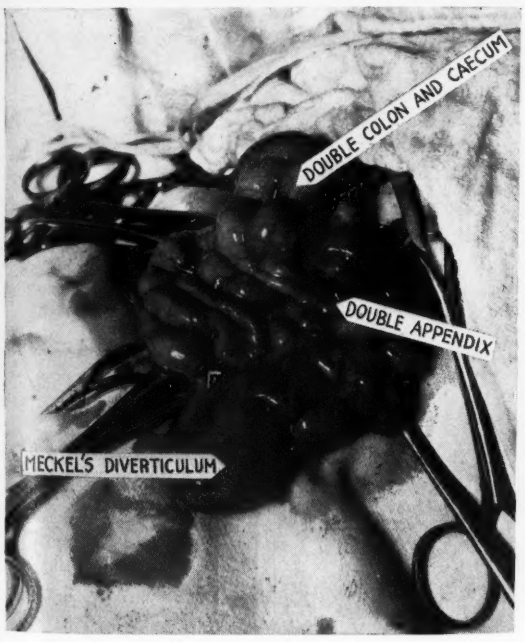

FIG. 467.—Appearance of the bowel at operation.

FIG. 468.—The sac of bowel (Meckel's diverticulum), showing the three issuing loops of ileum. X shows the entrance loop of ileum.

THE EFFECT OF TETRA-ETHYL AMMONIUM BROMIDE ON THE RENAL PELVIS AND URETER

A CONTRIBUTION TO THE STUDY OF HYDRONEPHROSIS

By A. G. MARSHALL

PATHOLOGIST, DUDLEY AND STOURBRIDGE AND DISTRICT HOSPITAL GROUP

THE drug tetra-ethyl ammonium bromide (T.E.A.B.) is believed to cause blockage of the ganglia of the autonomic nervous system, both sympathetic and parasympathetic (Lyons and others, 1947). There seems to be little risk when it is given by the intramuscular route, even in severe hypertension, but intravenous use is not so safe.

In the course of an investigation into the relationship between pyelonephritis and hypertension, a series of intravenous pyelograms was obtained in which there was a mild hydrocalix or dilatation of the renal pelvis. In some instances a constriction appeared to be present, and in order to determine if this was the result of muscular spasm, T.E.A.B. was administered by intramuscular injection and the pyelograms were repeated.

The results so obtained are thought to be sufficiently interesting to warrant publication so that other workers with better facilities may apply this method to the investigation of ' congenital ' hydronephrosis.

Technical.—Six consecutive cases of essential hypertension were investigated. The only selection was on the basis of an early age of onset, the youngest being 18, the oldest 41.

After physical examination and pathological investigation, an excretory pyelogram was taken. Each was thought to show some abnormality, though there is some doubt in *Case* 6. After a few days' rest, T.E.A.B. was given intramuscularly in doses ranging from 0·6 g. to 0·75 g. The intravenous injection for the pyelogram was given immediately afterwards and blood-pressure readings were taken at 5-minute intervals. The maximum effect of the T.E.A.B. appears between 15 and 30 minutes after the injection, and the optimum times for the X-ray photographs were estimated in each case from the wet films.

In *Case* 1 the T.E.A.B. was given intravenously and the patient experienced a mild vascular collapse.

CASE REPORTS

Case 1.—Sheila B., aged 18. Headaches and giddy turns for four years. No papillœdema. B.P. 145/110 mm. Hg. Urine: trace albumin; deposit, no cells. Blood-urea 44 mg. per cent.

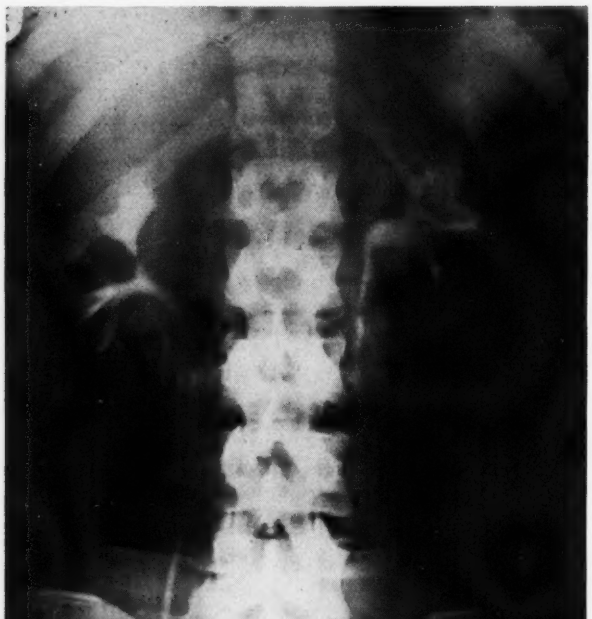

FIG. 469.—*Case* 1. I.V.P. 15 min.

FIG. 470.—*Case* 1. I.V.P. 14 min. T.E.A.B. (intravenous) 6 min.

The pyelograms (*Figs.* 469, 470) show a dilated upper calix with a narrowed infundibulum on the right side, and this has persisted in three such examinations over the last six months.

After T.E.A.B. this infundibulum is much wider and there is a slight dilatation of the whole pelvis. There is no marked change on the other side. The B.P. fell to 120/90 mm. Hg.

Case 2.—Dorothy B., aged 26. Attacks of headache and vomiting for eight years. B.P. 160/105 mm. Hg.

FIG. 471.—*Case* 2. I.V.P. 20 min.

FIG. 472.—*Case* 2. I.V.P. 25 min. T.E.A.B. 30 min.

Papillœdema present. Urine, trace albumin, but no cells nor casts in the deposit.

I.V.P. (*Figs.* 471, 472) showed a distorted pelvis of the right kidney. With T.E.A.B. there is some relaxation of the upper calix. There is no change on the other side. The B.P. fell to 110/75 mm. Hg.

Case 3.—Edith D., aged 28, had a swelling of the face and ankles during her third pregnancy but no record of her B.P. nor urine examination could be obtained. Now, a year after parturition, she has had a recurrence of the same symptoms. Her B.P. is 130/90. The fundi are normal and the urine contains a 'cloud' of albumin with R.B.C. and W.B.C. in the deposit.

I.V.P. (*Figs.* 473, 474): There is a sharply defined ring at the right pelvi-ureteral junction, with an elongated dilated ureter and a slight dilatation of the pelvis. This ring was present in each film of the series so that they were superimposable.

With T.E.A.B. the first film showed the ring to be in exactly the same position. Later there was considerable relaxation at this point, with a more normal shape. The left kidney showed no change.

Case 4.—George H., aged 41. After two weeks' diarrhœa and stomatitis he was found to have some swelling of the ankles. His B.P. was 140/90 mm. Hg; his urine contained a 'heavy cloud' of albumin. Red cells and casts were found in the deposit; the blood-urea was 220 mg. per cent. He was transferred from another hospital as dying of chronic nephritis, but showed considerable improvement on symptomatic treatment. No papillœdema was present. On discharge B.P. was 140/100.

I.V.P. showed an elongated dilated right pelvis with narrow calices. With T.E.A.B. the change is slight, but there is a return to normal of the pelvi-ureteral junction. The other side is not visualized. (*Figs.* 474, 475.)

Case 5.—Frederick C., aged 40. Six months ago severe epistaxis. Admitted to hospital in second attack. B.P. 230/150 mm. Hg. No papillœdema. Urine, light cloud albumin with occasional R.B.C., W.B.C., and casts. Sterile.

Pyelogram (*Figs.* 477, 478): Bilateral hydronephrosis and hydro-ureter, more marked on the right, where the ureter is elongated and folded in its central part.

With T.E.A.B. there is a marked change in both pelves and ureters, and disappearance of the fold.

Case 6.—Cyril L., aged 23. Complained of double vision. He gave a history of 'nephritis' while serving in the Army and was in hospital, where he was given "tablets". He thinks he was febrile and he had increased frequency but no hæmaturia.

I.V.P. was thought to show a mild hydronephrosis on the right side and the left side was not adequately visualized.

With T.E.A.B. there was no change. (*Figs.* 479, 480.)

FIG. 473.—*Case* 3. I.V.P. 15 min.

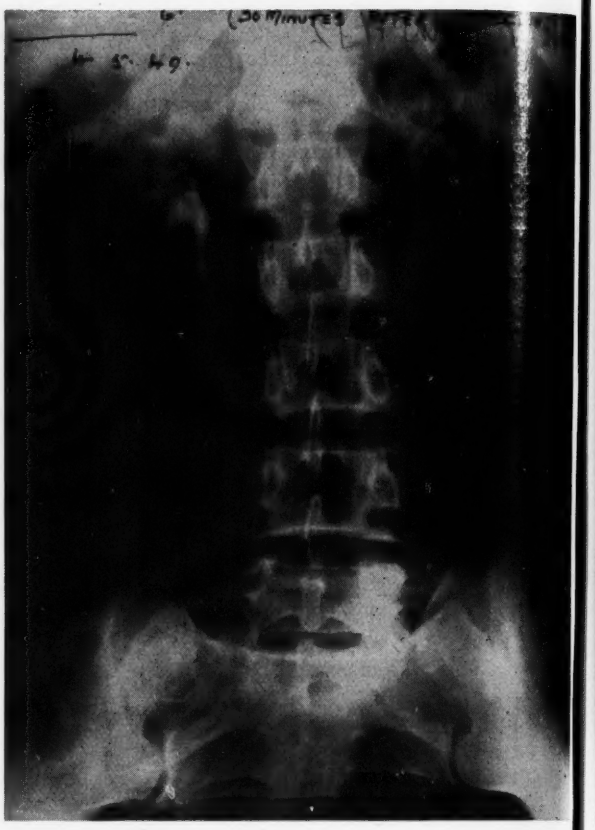

FIG. 474.—*Case* 3. I.V.P. 25 min. T.E.A.B. 30 min.

FIG. 475.—*Case* 4. I.V.P. 15 min.

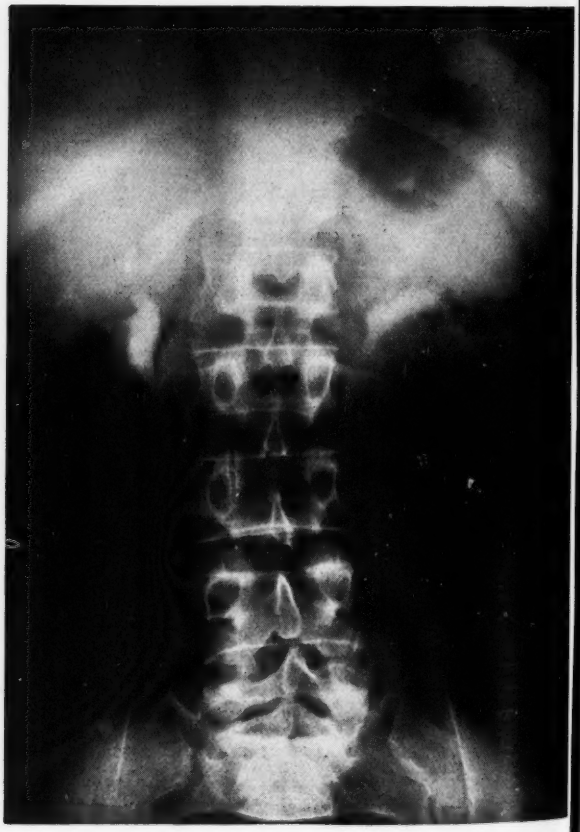

FIG. 476.—*Case* 4. I.V.P. 15 min. T.E.A.B. 20 min.

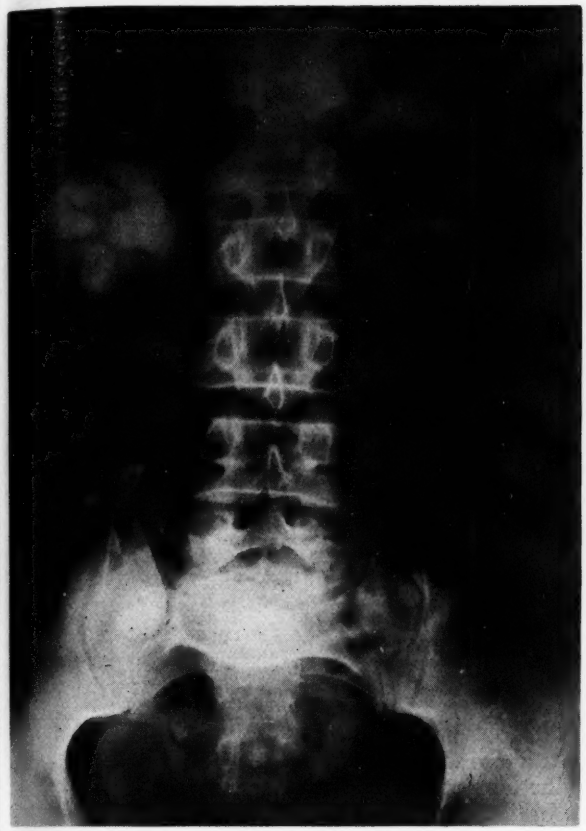

FIG. 477.—*Case* 5. I.V.P. 45 min.

FIG. 478.—*Case* 5. I.V.P. 40 min. T.E.A.B. 45 min.

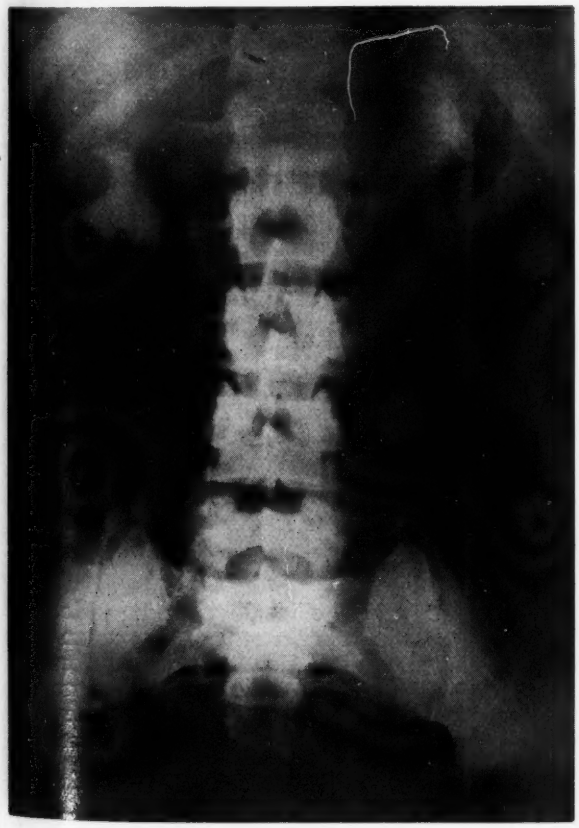

FIG. 479.—*Case* 6. I.V.P. 10 min.

FIG. 480.—*Case* 6. I.V.P. 25 min. T.E.A.B. 30 min.

DISCUSSION

The effect of T.E.A.B. on the renal pelvis and ureter in the first five cases seems to be a return towards normal. Constrictions disappear, the pelvi-ureteral junction assumes a smoother funnel-shaped outline, dilated ureters tend to a more normal diameter, and bends and kinks in them seem to be reduced.

In *Case* 6 the failure to show any change is open to two explanations. As suggested above, this pyelogram may be within normal limits; or, alternatively, it is probable that he had pyelitis in the Army and the pelvis will no longer contract normally.

It remains to exclude the physiological changes described by Cumming (1931). The cases are consecutive and the photographs were from the only series in each case except the first. In several of the figures the shadow on the other side has not changed, and in *Case* 1 the infundibular constriction remained unaltered in three examinations over six months. Finally, in some of the cases there is evidence of resumption of the abnormality in the later films of the series.

The nerve-supply to the pelvis and ureter is described by Lowsley and Kirwin (1944) as sympathetic and parasympathetic. Samson Wright (1940) states that the sympathetic supplies both motor and inhibitory fibres. Satani (1919) described ganglion cells in the external ureteric coat. Learmonth (1931) found sympathetic stimulation caused constriction of the ureteric orifices, which he thought was independent of the contraction of the trigone. Kelly and Burnam (1914) appear to have been the first to describe localized thickenings of the muscle coat at the infundibula, the pelvi-ureteral junction, and in the ureter; it may be that these have a separate autonomic control.

As far as any conclusion can be drawn from so small a series there seems to be two possible explanations of the changes :—

a. Resumption of normal tone in a dilated structure. Underwood (1936) showed that the wall of a hydronephrosis is inert rather than distended and that it would contract with adequate stimulation. It is therefore possible that the effect of the drug has been the elimination of inhibitory impulses.

b. Relaxation of localized spasm. It is not extravagant to postulate such a condition in these cases, and it seems likely that autonomic paralysis might allow relaxation, with the result that intrinsic muscular tone may take control. The similarity of such a condition to that described by Bodian, Stephens, and Ward (1949) in Hirschsprung's disease would be striking, and indeed a combination of dilatation of the colon, ureters, and bladder has been described (Learmonth, 1935).

SUMMARY

1. Excretion pyelography has been performed on six patients after the administration of tetra-ethyl ammonium bromide.

2. As a result, early hydronephrosis and hydroureter tend to be abolished.

3. It is suggested that the drug might relax localized muscular spasm.

4. This report is submitted in the hope that the method may be found useful in investigating the cause of hydronephrosis.

I wish to thank Dr. A. P. Booth, in whose departments these investigations were made, for his help. Miss T. Lewis and other members of his staff have given me every assistance.

The patients are under the care of my colleagues on the Staffs of the Hospitals in this Group, and I am glad to acknowledge my indebtedness to them.

Professor G. Hadfield has kindly given me advice and help.

I am grateful to Mr. D. R. Paton, who made the reductions of the X-ray photographs.

REFERENCES

BODIAN, M., STEPHENS, F. D., and WARD, B. C. H., (1949), *Lancet*, **1**, 6.

CUMMING, R. E. (1931), *J. Urol.*, **25**, 613.

KELLY, H. A., and BURNAM, C. F. (1914), *Diseases of the Kidneys, Ureters and Bladder*, **1**, 169. New York.

LEARMONTH, J. R. (1931), *Brain*, **54**, 147.

— — (1935), *Surg. Gynec. Obstet.*, **60**, 114.

LOWSLEY, O. S., and KIRWIN, T. J. (1944), *Clinical Urology*, 2nd ed., **2**, 1212. Baltimore.

LYONS, R. H., MOE, G. K., NELIGH, R. B., HOOBLER, S. W., CAMPBELL, K. N., BERRY, R. L., and RENNICK, B. R. (1947), *Amer. J. med. Sci.*, **213**, 315.

SATANI, Y. (1919), *J. Urol.*, **3**, 247.

UNDERWOOD, W. E. (1936), *Jacksonian Prize Essay, Royal College of Surgeons*, **2**.

WRIGHT, SAMSON (1940), *Applied Physiology*, 7th ed., 177. London.

DUPLICATIONS OF THE ALIMENTARY TRACT

BY G. H. WOOLER

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DUPLICATIONS of the alimentary tract are not common, and except for Ladd and Gross's (1947) personal series of 18 cases, few have been described. They may be of a spherical or tubular shape. The tubular duplications have been recognized for a long time, but many of the spherical ones have received other names in the literature, such as enterogenous cysts, enteric cysts, inclusion cysts, and giant diverticula—thus hiding their true nature.

Persistence of the vitelline duct causing a Meckel's diverticulum occurs in 2 per cent of the human race. Other minute diverticula develop as buds from the epithelium lining the intestine and grow into the subepithelial connective tissue, where they lie chiefly on the antimesenteric side of the intestine—in the same position as Meckel's diverticulum.

Duplications of the alimentary tract may occur anywhere from the mouth to the anal canal, but they

are found most commonly in relation to the small intestine. Here they lie between the layers of the mesentery on the mesenteric side of the intestine.

FIG. 481.—The duplication after it had been opened. It shows the two peptic ulcers in the jejunum and the gastric mucosa lining the duplication.

Their walls contain all the layers of the normal intestine, and are therefore thick and muscular. They cannot be separated easily from the adjoining intestine. It is important to realize this fact at operation, for it distinguishes them from mesenteric cysts. The latter also lie between the layers of the mesentery, but being of lymphatic origin they have a thin wall and are easily separable from the adjacent intestine.

Embryology. — The formation of duplications is not really understood and many theories have been suggested, of which I should like to mention two :—

1. An epithelial bud develops from the intestinal mucosa and grows into the loose submucosa. It then forms a cyst which finds its way through the circular muscle along one of the openings through which the vessels supply the mucosa. It spreads in the intermuscular tissue or may finally pierce the longitudinal muscle as well. (Bremer, 1944.)

2. In an embryo six weeks old the intestinal epithelium proliferates to such an extent that the lumen may be occluded. The cells of the thickened epithelium secrete a fluid which collects into vacuoles. These are arranged in rows longitudinally along the intestine (Johnson, 1910). The vacuoles finally open into the lumen as the intestine grows in size. In rare cases the vacuoles retain their form and become intestinal duplications.

CASE REPORT

A female child aged 8 months was first admitted to The London Hospital in 1937 with increasing pallor and melæna. Hæmoglobin 19 per cent ; red cells 2,000,000 ; normal white-cell and differential count on admission. These were the only abnormal physical signs, and as there was no obvious cause for the melæna she was kept under observation. She did not improve and the melæna continued, so laparotomy was performed. A small diverticulum of the jejunum was found in the left iliac fossa surrounded by numerous adhesions. There were many yellow-grey nodules in the mesentery and great omentum. She was thought to have a tuberculous peritonitis, and part of the greater omentum was removed for biopsy. Microscopical section of the omentum showed " epithelioid and giant-celled foreign-body reaction around small areas of homogeneous coagulation ". She was given repeated blood transfusions, recovered completely, and remained well without any symptoms for the subsequent eight years.

In 1946, one week before her second admission to hospital, she vomited about an hour after her tea. The following day she had lower abdominal pain which doubled her up, and she was constipated. Four days before admission she had diarrhœa and passed five stools, coal-black in colour. She took only a fluid diet, but the lower abdominal pain persisted.

ON ADMISSION.—She was a pale, underdeveloped and undernourished child. The abdomen showed visible peristalsis passing from the right hypochondrium to the left iliac fossa. A firm sausage-shaped mass was palpated in the left iliac fossa and there was generalized lower abdominal tenderness. Hæmoglobin was 81 per cent.

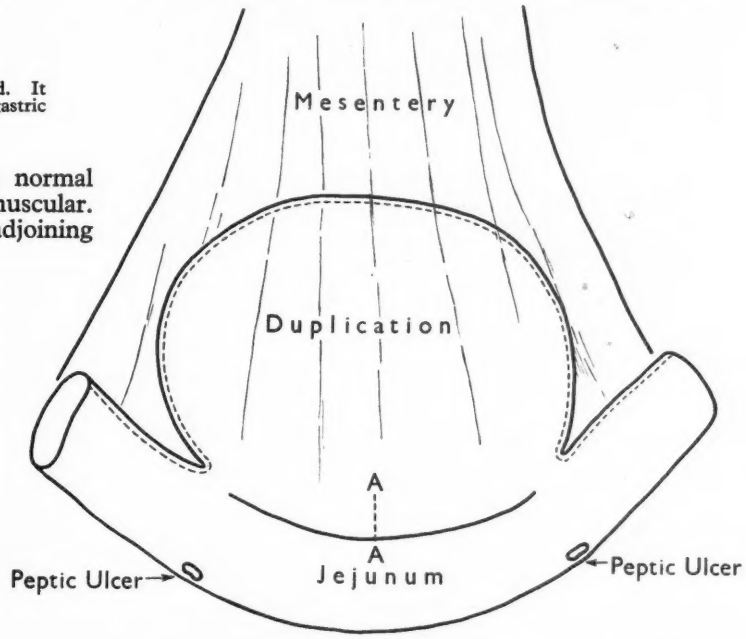

FIG. 482.—The duplication, which was lying between the layers of the mesentery, communicated with the lumen of the jejunum in two places. Opposite these communications were peptic ulcers in the jejunal wall. The specimen was opened by dividing along the dotted line.

AT OPERATION.—Laparotomy was performed through a left paramedian incision. The jejunum was dilated, the ileum empty. As the dilated jejunum was followed, it led to a mass of inflammatory adhesions in the left iliac fossa between the small intestine and the omentum. They covered a cystic swelling which was lying in the

mesentery. After the adhesions had been dissected away, a perforation was found in the jejunal wall close to the swelling. At this point the jejunum was kinked and obstructed. The affected part of the jejunum was resected and an end-to-end anastomosis performed.

The child made an uninterrupted recovery and within two months of her discharge from hospital had gained 1 st. in weight. She had never looked or been so well in her life.

PATHOLOGY.—The specimen consisted of 14 cm. of jejunum, to the mesenteric side of which the duplication

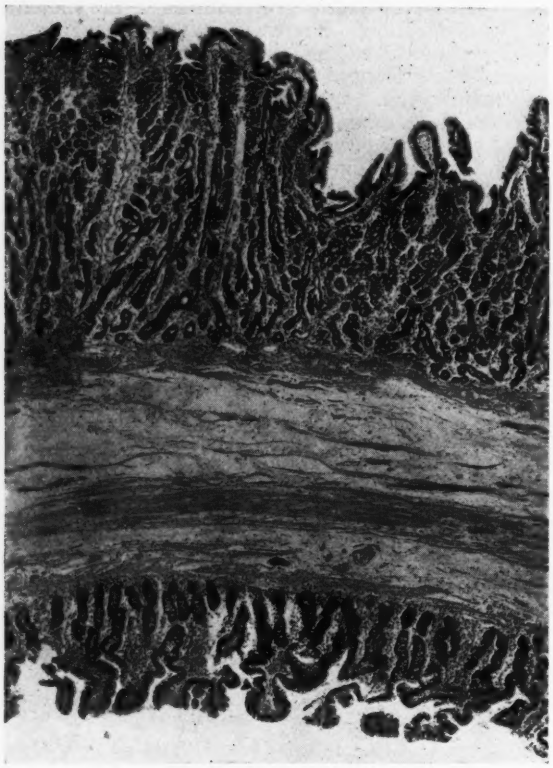

FIG. 483.—Section taken across the wall (A–A in *Fig.* 482) separating the duplication from the jejunum. It shows the hypertrophic gastric mucosa on the one side and jejunal mucosa on the other. (*See* histological report.)

was attached. This was a circular hollow swelling, approximately 9 cm. in diameter. The cavity of the duplication communicated with the lumen of the jejunum at the proximal and distal ends of its attachment to the jejunum; between these openings it was separated from the jejunum by a septum. Peptic ulceration of the jejunum had occurred opposite the openings of the duplication. One of these ulcers had perforated, but leakage had been prevented by adherent omentum. Bleeding from these ulcers had caused the severe melæna.

The duplication had its own blood-supply and some vessels coursed over its surface to supply the adjoining jejunum.

Histology (Dr. C. J. E. Wright).—The duplicated portion is lined by a thick layer of gastric fundal-type mucosa with typical glands showing chief cells and parietal (oxyntic) cells. The mucosal stroma is heavily infiltrated by lymphocytes, plasma cells, and eosinophils, with smaller numbers of polymorphs and histiocytes. The mucosa of the small intestine proper also shows heavy lymphocytic, plasma-cell, and eosinophil infiltration. The small bowel and duplicated bowel together are surrounded by a thick main muscular coat formed of the usual inner circular and outer longitudinal layers. Where the two portions of bowel are adjacent the mucosæ are in close proximity, being only separated by loose connective tissue in which there is a much attenuated and broken up muscularis, completely absent over wide areas, and continuous with the inner muscle coat surrounding the two portions; the muscularis mucosa is well-defined in both mucosæ. There is an acute fibrinous peritonitis.

Moll (1926) had a similar case to this one. The duplication was 33½ in. long, tubular in shape, and was lying along the mesenteric attachment of the ileum. It was lined by gastric mucosa. The specimen is now under Professor Stewart's care in the Pathology Museum at Leeds. Unfortunately, it was not histologized before publication.

SUMMARY

A rare cause of severe melæna occurring in a female infant of 8 months is described. The true nature of the condition was not diagnosed until she was 8 years old, when she presented with acute intestinal obstruction and melæna.

The cause was a duplication of the jejunum lined with gastric mucosa. This had produced peptic ulceration of the adjacent jejunum. The ulcers had bled, and one had perforated, but the perforation had been walled off with adhesions which had caused acute intestinal obstruction.

Resection of the duplication with the adjacent jejunum was carried out, and an end-to-end anastomosis performed. The child's health immediately improved so rapidly that her relations did not recognize her when she returned from a month's convalescence.

I am grateful to my colleague Dr. Maxwell Telling for his helpful criticism. I wish to thank Professor Turnbull for examining the specimen for me and for his report of the histology of the omentum. I should also like to pay tribute to my former friend and colleague, the late Keith Riddoch, who was working with me at the time and was actually the first person to diagnose this particular case. His enthusiasm and friendship we all miss so much.

REFERENCES

BREMER, J. L. (1944), *Arch. Path.*, **38**, 132.
JOHNSON, F. P. (1910), *Amer. J. Anat.*, **10**, 521.
LADD, W. E., and GROSS, R. (1940), *Surg. Gynec. Obstet.*, **70**, 295.
— — — — (1947), *Abdominal Surgery of Infancy and Childhood.*
LEWIS, F. T., and THYNG, F. W. (1907), *Amer. J. Anat.*, **7**, 505.
MOLL, H. H. (1926), *Brit. J. Surg.*, **14**, 176.

SHORT NOTES OF RARE OR OBSCURE CASES

A CASE OF OSTEOCHONDRITIS DISSECANS OF THE ANKLE

By T. R. SIMPSON, Salisbury, S. Rhodesia

While osteochondritis dissecans of the knee is comparatively common, the occurrence of the condition in the ankle appears to be sufficiently unusual to warrant the recording of a single case.

CASE REPORT

History.—Miss Z. M., a European schoolteacher, aged 26, was first seen on July 16, 1948, when she stated that she had " put out " her left ankle a few hours previously. She had been sitting on the floor in a cross-leg position, and on rising to her feet she had felt a sudden sharp pain in the left ankle, and immediately had the sensation that the joint was locked. The pain had rapidly subsided to a dull ache, but the sensation of locking persisted.

She gave a previous history of a severe sprain of the left ankle in 1940, since when there had been occasional brief, sharp pains in the joint, and on one occasion twelve months ago momentary locking had occurred. Those subsequent incidents were not provoked by any further particular trauma to the ankle.

On Examination.—The left ankle-joint was held in a position of mid-plantar flexion. The only active movement possible was a slight increase in the amount of plantar flexion. On passive movement a greater degree of plantar flexion was obtained, lateral movements at the subtaloid joint were present, but dorsiflexion of the foot appeared to be solidly blocked and the attempt to produce it caused considerable pain within the ankle. There was no obvious swelling of the joint. A localized area of tenderness was present on the anterolateral aspect at the level of the joint line.

Radiological examination (*Fig.* 484) revealed a loose body, apparently completely separated and lying in the

A B

Fig. 484.—Showing loose body in the joint and rarefied area on superolateral surface of talus.

posterolateral part of the ankle-joint. Although the site of origin of this loose body could not be clearly defined in the radiographs, there was a suggestion of rarefaction of a small area on the lateral edge of the superior articular surface of the talus. On the anterior margin of the lower articular surface of the tibia there was seen a small osteophytic outgrowth, which appeared firmly fixed at its base and showed no evidence of recent injury.

A tentative diagnosis of osteochondritis dissecans was made and operation for removal of the loose body was performed on July 26, 1948.

At Operation.—On exploration of the joint it was found that the osteophyte on the tibia did not impinge

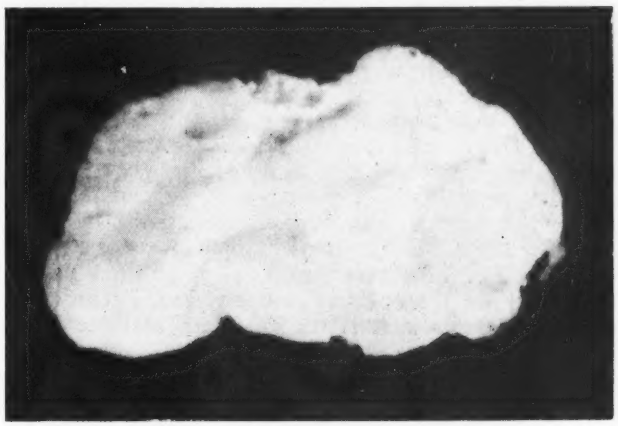

Fig. 485.—Loose body removed at operation. (× 2·5.)

upon the talus at any point and could not be responsible for any mechanical interference with dorsiflexion of the foot, nor could it have caused sudden locking of the joint. The superior articular surface of the talus was examined and on its lateral edge, at about the summit of its convexity, there was seen an area approximately 1 cm. long by 0·5 cm. wide which lacked normal articular cartilage. The surface of this area did not glisten, it was rough in appearance, yellowish in colour, and its texture was soft. It was very slightly depressed below the level of the surrounding normal articular cartilage. Further inspection revealed a loose body lying in the posterolateral part of the joint. It was firmly impacted between the articular surfaces of the tibia and talus, but was removed without difficulty, whereupon full passive dorsiflexion of the foot was obtained. The defect on the articular surface of the talus was not interfered with, as it appeared that a certain amount of repair had already taken place, and it was thought inadvisable to interrupt this process.

The loose body (*Fig.* 485) appeared to consist of bone with patches of smooth cartilage on its surface. It measured 12 × 6 × 3 mm., and, judging by its size and slightly curved shape, it could have originated from the area noted on the talus. No other possible site of origin was found in the joint. The wound was closed in layers and a light plaster applied to support the foot. Recovery was uneventful except for a mild superficial wound infection which rapidly subsided after the extrusion of a piece of catgut.

Progress.—The patient was seen again on Nov. 23. She was then fully active in her work and in games, and felt no disability in the ankle. There was free and painless movement of the ankle, except that the terminal few degrees of full dorsiflexion were absent.

COMMENT

So far as can be ascertained from the limited literature available to the writer, only about 50 cases of osteochondritis dissecans of the ankle have been reported.

Mensor and Melody (1941), in a review of the literature, found 19 cases and added 1 case of their own. Hutchison (1943) reported 1 case, and Vaughan and Stapleton (1947) described 4 cases. Ray and Coughlin (1947) published a series of 13 cases, in one of which both ankles were affected. The last writers refer also to 3 further cases discovered by personal communication, to 1 reported by Schnaberth in 1939, and to four papers, recording 9 cases, which appeared between 1942 and 1944, but which are not available to me.

The case here reported is interesting in several respects. The history of definite and fairly severe trauma eight years ago, followed by intermittent pains in the joint for a considerable period before the development of the state of affairs found at operation, lends some support to the traumatic theory of origin of the condition.

The fact that the loose body had already separated when the patient was first seen is unusual (Vaughan and Stapleton) and can be taken to account for the somewhat atypical radiological appearance on the superior surface of the talus. There was seen only a suggestion of rarefaction of the affected area, instead of the usual picture of a clear line of demarcation overlying which is the fragment of bone and cartilage due to be cast off. In the present case the radiograph could be interpreted as showing partial repair of the defect left after separation of the loose body.

At operation, also, the appearance was that the defect in the talus had, to some extent, already been repaired by fibrous tissue and new bone. Certainly the depth of the crater remaining at the time of operation was insufficient to accommodate the full thickness of the loose body.

The time at which actual separation of the loose body took place is difficult to judge. It may have been at or before the occasion of momentary locking of the joint one year ago. Alternatively, only a partial separation may have occurred at that time, with complete separation at the time of the incident for which operation was performed. The radiological and operative findings would appear to support the former contention.

I wish to thank the Medical Superintendent, Salisbury General Hospital, for facilities granted to me in treating this case, and I am indebted to Dr. N. Daniels for the radiographs.

REFERENCES

HUTCHISON, R. G. (1943), *Brit. J. Radiol.*, **16**, 147.
MENSOR, M. C., and MELODY, G. F. (1941), *J. Bone Jt. Surg.*, **23**, 903.
RAY, R. B., and COUGHLIN, E. J. (1947), *Ibid.*, **29**, 697.
VAUGHAN, C. E., and STAPLETON, J. C. (1947), *Radiology*, **49**, 72.

A CASE OF TUBERCULOSIS OF THE COLON WITH CARCINOMA OF THE RECTUM

BY PAUL HICKINBOTHAM

TUBERCULOUS disease of the colon, other than the hypertrophic ileocæcal form, is uncommon; multiple tuberculous ulcers sometimes accompany those found in the small bowel in patients who are subjects of open pulmonary tuberculosis, but this is unusual. The case now reported is therefore of interest in that it demonstrates a very chronic stenosing series of lesions of the large bowel, associated with pulmonary tuberculosis, and terminating fatally following the development of a carcinoma of the rectum.

CASE REPORT

Mr. W. K., aged 32, was admitted to hospital on Sept. 17, 1936, with a history of intermittent attacks of severe colicky pain in the right iliac fossa, sometimes associated with vomiting; during attacks he generally had slight diarrhœa. Otherwise he was in good health. On examination he was afebrile but definitely tender in the right iliac fossa; rectal examination revealed nothing abnormal, and there is no record of the state of his chest.

Laparotomy performed the following day revealed a distended œdematous cæcum, with a very œdematous but not inflamed appendix lying behind it. A mass was felt in the ascending colon with peritoneal puckering over it and there was a similar mass in the transverse colon; these were considered to be of a subacute inflammatory nature and were short-circuited by an ileotransverse anastomosis. His convalescence was satisfactory.

He was readmitted for investigation two months later; he had put on weight and was apparently in excellent condition. The Wassermann reaction was negative; occult blood was present in his stools; radiological examination after a barium enema showed the ileotransverse anastomosis to be functioning well, but there was a filling defect in the transverse colon and an early stricture of the pelvic colon, suggestive of tuberculous disease. Examination of the stools for tubercle bacilli was apparently done, but there is no record of the result.

Following this he was discharged; when seen three months later he was very well, having gained further weight and having a good appetite. He failed to keep an appointment a further three months later, and was not seen until about twelve years later when he was admitted on Feb. 8, 1949.

In the intervening years he had had some lower abdominal pain, worse after meals, but never bad enough to bring him to hospital. For the last six months, however, he had had alternating diarrhœa and constipation with considerable loss of weight, and for the last ten days he had had some lower abdominal colic.

On examination he had clearly lost a lot of weight; he had a large growth of his rectum, biopsy of which showed it to be an anaplastic carcinoma. X-ray examination of his chest revealed calcified lesions in the right upper zone, with infiltration—possibly active—in the left upper zone. A barium enema showed a constriction 1½ in. long in the sigmoid colon and a further

narrowing in the transverse colon, beyond which no barium would flow.

After correction of his anæmia by transfusion, operation was performed on Feb. 18. The rectal carcinoma was found to be operable, with no secondaries palpable; a narrowing was present in the pelvic colon, another in the ascending colon, and the previous ileo-transverse anastomosis was involved in an inflammatory stenosing process producing partial obstruction. Synchronous combined excision of the rectum was performed, the sigmoid stricture being included in the specimen (*Fig.* 486). The old anastomosis was finally enlarged by a plastic procedure.

FIG. 486.—Photograph of operation specimen, showing tuberculous stricture of sigmoid colon and carcinoma of rectum.

His post-operative course was stormy, and the lower part of his abdominal wound broke down, with the subsequent development of a fæcal fistula. On March 15, a month after operation, his wound was resutured and the fistula closed. His condition, however, steadily deteriorated and he died two days later.

AT AUTOPSY.—Autopsy revealed the following findings: chest, bilateral basal pneumonic consolidation, with active tuberculous cavitation at the left apex and healed lesions at the right apex. The colon from the ileocæcal junction to 4 in. beyond the ileotransverse anastomosis was grossly diseased, with thickening of the wall, multiple small mucosal ulcers, small subserous tubercles, and almost complete stenosis at three points. Histology of the operation specimen showed a typical tuberculous ulcer of the sigmoid and anaplastic carcinoma infiltrating the bowel wall but not invading the glands (*Figs.* 487, 488). Sections of the autopsy specimen showed chronic inflammatory ulceration of the colon with tuberculous lesions in the paracolic glands.

COMMENTARY

Simultaneous occurrence of tuberculous and malignant disease in the same lesion in the colon has been reported by several authors (Randall and Spalding, 1946): in such cases there is always discussion as to the nature of the association of the

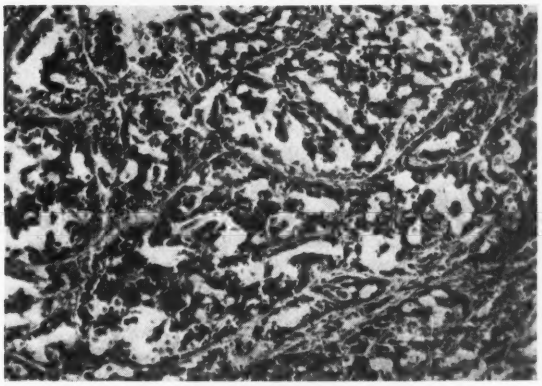

FIG. 487.—Section of tuberculous stricture of sigmoid. (× 60.)

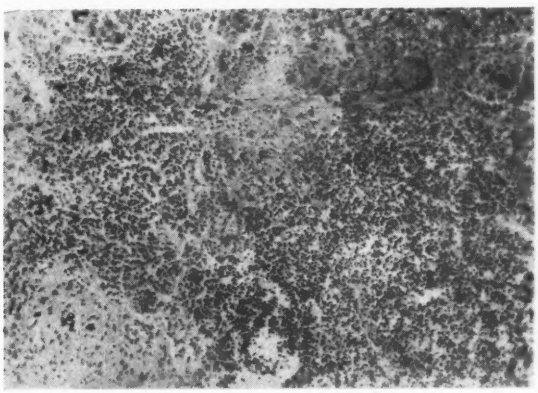

FIG. 488.—Section of carcinoma of rectum. (× 60.)

two conditions. In the case above, however, the two conditions were present in different parts of the colon, so that there can hardly be any question of association. The main interest of the tuberculous lesions in the case lies in their extreme chronicity—a relatively slight advance in the stenosing process over a period of twelve years.

My thanks are due to Mr. Donald McGavin, under whom this patient was admitted to hospital, for permission to report this case.

REFERENCE

RANDALL, K. J., and SPALDING, J. E. (1946), *Brit. J. Surg.*, **33**, 372.

A CASE OF TRAUMATIC AVULSION OF A MAIN-STEM BRONCHUS FROM ITS LUNG, TREATED BY IMMEDIATE PNEUMONECTOMY

By E. R. G. KIRKPATRICK, Perth

The tearing or rupture of a main-stem bronchus is a rare injury, only 33 such cases having been recorded in the English literature since 1889, so far as the author's search has disclosed. It is probable that other cases have occurred, but have not been recorded. Owing to its severe nature, and its frequent association with other injuries, the diagnosis of this injury may fail to be made and the victims often succumb shortly after the accident. The case described below is put on record because of the rarity of the injury, and because it is felt that, if such an injury is diagnosed or suspected, an exploration of the chest, and, if necessary, bronchial suture or pneumonectomy, may be the means of saving the patient's life, or much subsequent morbidity should the patient recover, as it seems that *all* such cases develop bronchial occlusion and pulmonary collapse.

It appears that no case of such an injury has had the thorax opened on the same day as the injury, prior to the one reported here, so a description is given of the appearances of the torn bronchus shortly after the accident.

Hodes, Johnson, and Atkins (1948), in describing post-traumatic bronchial occlusion with pulmonary collapse, have collected 33 cases from the literature, including 1 case of their own. Of these, 11 survived the immediate effects of the injury, and all developed bronchial occlusion. Four of the cases which recovered subsequently required removal of the collapsed lung at varying intervals from five months to twenty-three years after the accident, for symptoms due to mediastinal shift or infection; of the remaining 7 cases, 2 were cases in which the injury was discovered at post-mortem twenty-one and twenty-three years later; 3 were apparently well, and the other 2 had collapsed lungs with varying disability. The injury in all cases described was close to, or within a few centimetres of, the tracheal bifurcation, affecting the right and left side more or less equally. Practically all were the result of the chest being crushed by a vehicle, generally being run over by a wheel. The mechanism of the injury by which the lesion is produced can be only conjectured. It is believed by the author that when the chest is crushed, with the glottis closed, the two air-filled lungs are pushed apart by the approach of the sternum to the vertebral column. One or other lung is thus torn, in varying degree, from its bronchus. In some of the cases reported the thoracic cage has escaped obvious damage.

Kinsella and Johnsrud (1947), in a comprehensive review of this injury, report finding 38 cases, 19 of which recovered, described in the literature, including 2 of their own, since the first case of recovery recorded by Kritinski in 1928. Of these they say that 10 cases may be classed as spontaneous recoveries, and the others required active treatment, such as decompression of tension pneumothorax, suprasternal incision for mediastinal emphysema, etc. They recommend that due consideration should be given to active surgical intervention, if the patient's general condition will permit, and that, if possible, the torn bronchus should be sutured when incompletely torn, but if the bronchial artery, which is nutrient to the lung, be interrupted, excision of the lung or lobe involved is called for. They consider that the risk of an exploration might prove very slight with modern anæsthesia, blood banks, and modern technique in thoracic surgery, as compared with doing nothing at all.

CASE REPORT

J. C. H., a boy 13 years of age, was admitted to hospital at about 10.30 a.m. on Oct. 25, 1948, in a very shocked and distressed condition, following an accident approximately an hour previously, in which he had been run over by the wheel of a farm tractor trailer. When examined shortly after admission he was found to have signs of right-sided pneumothorax, and a mediastinal shift to the left, surgical emphysema of neck and thoracic wall, a 4-in. lacerated wound of all layers of the scalp over the left parietal region, and a fractured left clavicle. He was extremely distressed, considerably cyanosed, restless, and anxious looking. His pulse-rate was 152, and his respiration rate about 55 per minute. A speedy aspiration of his right pleural cavity liberated air, apparently under pressure, with considerable relief to the patient. On withdrawal of the aspirating needle, his distress returned, so an intercostal cannula was inserted, and when this was connected to a water trap, he was found to have a positive inspiratory and a negative expiratory pressure in the right pleural cavity. He was treated for shock for the next three hours, during which time his general condition improved a little, the surgical emphysema increased somewhat, he had several distressing attacks of hæmoptysis, though none were large ones, but it was plain that something had to be done to close an obvious large bronchopleural fistula if he were to live.

At 2.30 p.m. the same day he was taken to the operating theatre, anæsthesia was induced and maintained by endotracheal cyclopropane, and the right pleural cavity apened by an incision in the sixth intercostal space. The right lung was seen to be collapsed and apparently uninjured. Respiratory air was seen to be issuing from a tear in the rather emphysematous upper mediastinum at the upper margin of the lung root; from this opening the right main bronchus was picked up and was found to be torn across completely just distal to its epiarterial branch, having been completely avulsed from its lung. It had then retracted into the mediastinum. A short length of the epiarterial branch, about 1 cm., remained attached to the proximal main stem, and the main bronchus was torn across irregularly at the level of this upper-lobe branch. There was about an ounce or so of blood in the pleural cavity, and the main vessels to the lung were undamaged. As the bronchus was torn so irregularly and completely with this small length of its epiarterial branch remaining attached to the main stem, it was considered that there was no possibility of suturing it, so the main stem was clamped and then trimmed and closed by silk sutures close to the carina. This left a lung with no hope of future aeration, so the main pulmonary artery and veins were dissected at the lung hilum

and divided between ligatures, and the lung was removed. It was noted that, once the bronchus was clamped, the patient's general condition and his breathing, of course, greatly improved.

Convalescence was more or less uneventful. He was treated in an oxygen tent for a few days, and systemic penicillin was given as well as intrapleural penicillin, with aspiration of the pleural exudate. The first radiographs of his chest, taken on Nov. 3, showed, in addition to the hydropneumothorax on the right side, the fracture of his left clavicle and transverse fractures of the anterior ends of the third and fourth ribs on the left side—the side opposite to the torn bronchus. He was discharged from hospital, healed and well, on Nov. 20. When seen on Jan. 12, 1949, he was still well, and radiography of his chest showed that his right pleural cavity was becoming obliterated. He had no complaints, and was taking moderate exercise without dyspnœa.

REFERENCES

HODES, P. J., JOHNSON, J., and ATKINS, J. P. (1948), *Amer. J. Roentgenol.*, **60**, 448.

KINSELLA, T. J., and JOHNSRUD, L. W. (1947), *J. thorac. Surg.*, **16**, 571.

KRITINSKI, S. I. (1928), *Virchows Arch.*, **266**, 815.

MECKEL'S DIVERTICULUM WITH A PERITONEAL CAUL

A UNIQUE CAUSE OF INTESTINAL OBSTRUCTION

BY J. F. CURR, EDINBURGH

THERE are several well-recognized methods by which a Meckel's diverticulum may give rise to intestinal obstruction, but the following case illustrates a mechanism which appears to be unique.

CASE REPORT

HISTORY.—A boy, aged 6 years, previously in the best of health, was admitted to the children's wing of Leith Hospital one Monday evening in 1945. Since the preceding Thursday he had complained of severe abdominal pain, the bowels had not acted, and vomiting had become continued and copious.

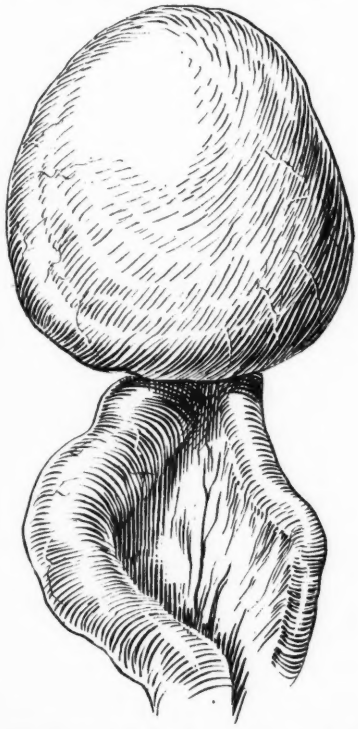

FIG. 489.—The original finding at operation—a ball-like mass into which a loop of ileum disappeared.

ON EXAMINATION.—The boy was dehydrated and the general condition was not good. There was a gross degree of abdominal distension, with moderate generalized tenderness and some free fluid in the peritoneal cavity. Rectal examination was negative and there was no blood-stained mucus; a soap-and-water enema produced no result. An obvious diagnosis of intestinal obstruction was made and the necessary administration of intravenous saline was commenced.

AT OPERATION.—Two hours later, under open ether anæsthesia, a right paramedian incision was made, two-thirds of it below the umbilicus, and on opening the peritoneal cavity a large quantity of straw-coloured fluid escaped. A freely mobile, tense, ball-like mass, about 3 in. in diameter, was palpated in the right iliac fossa and was easily delivered into the wound. The opaque covering of this mass took the form of a most peculiar caul or sac of peritoneal tissue and at one part of it there was a narrow opening into which a loop of bowel disappeared completely out of sight. The lower ileum was quite collapsed and there was great distension of the upper ileum and jejunum (*Fig.* 489).

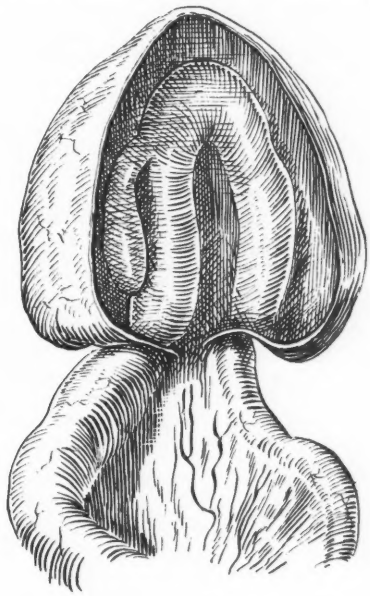

FIG. 490.—The mass opened by incising the caul and the contents exposed.

A constricting ring was slit open, allowing the escape of thin sanguineous fluid, and the incision was continued through the peritoneal caul towards the opposite pole of the mass, thus exposing the contents (*Fig.* 490).

This step released an internal strangulation, consisting of a loop of ileum, 10 in. long, with a Meckel's diverticulum, 2 in. long, attached to the antimesenteric border.

The peritoneal caul, when opened out completely, now assumed a fan-like shape (*Fig.* 491). It was attached to the antimesenteric border of the ileum for a distance of 6 in. and to both the proximal and distal aspects of the diverticulum, and it also extended as a fringe for 3 in. beyond the tip of the diverticulum. There was no

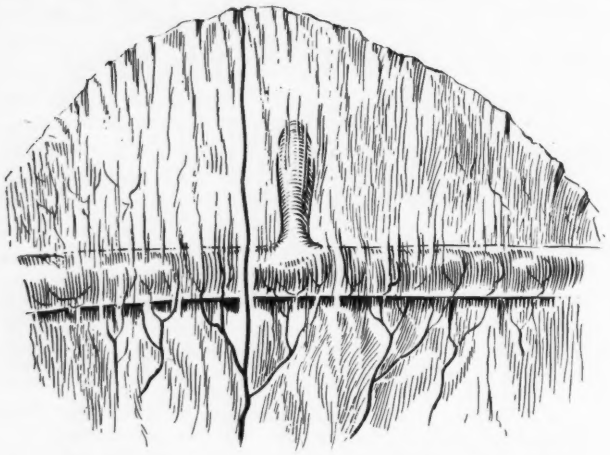

FIG. 491.—The abnormal mesentery laid open, showing its attachments to the ileum and the diverticulum, and the remnant of the vitelline artery.

connexion between the umbilicus and either the diverticulum or the caul, but the latter was continuous with the mesentery of the ileum by means of a filmy membrane passing over the right side of the ileum. A remnant of the vitelline artery arose from an artery in the mesentery, crossed over the right side of the ileum, passed along the proximal aspect of the diverticulum, and continued to the extremity of the peritoneal sac; it bled quite freely when divided and was ligated.

Once the strangulation had been relieved, the entire peritoneal caul was excised and the Meckel's diverticulum was resected. The strangulated loop of ileum recovered adequately and did not require resection.

PROGRESS.—For the next three days the general condition remained serious and the abdomen was even more distended than before operation. However, on the third day the bowels acted spontaneously and rapid improvement and a complete recovery followed.

DISCUSSION

The Anatomical Abnormality.—The abnormal peritoneal fold which formed the caul was no doubt in the nature of a distorted mesentery of the Meckel's diverticulum, and before the onset of intestinal strangulation the configuration appeared to have been as in *Fig.* 492. It spread out from its attachments to the ileum and the diverticulum and formed a large blind peritoneal sac, which was a potential site for strangulation, especially as its base was constricted in comparison to the expanded fundus. At the extremities of its attachments to the ileum, the short free margin of the caul held the afferent and efferent limbs close together, this being the part of the constriction ring cut through at operation.

The Mechanism of the Obstruction.—Strangulation eventually became established when the diverticulum and the loop of ileum rotated and were forced into the fundus of the peritoneal sac; the narrower neck of the sac became the constricting ring. It was in this way that the ensnared gut could not be seen on original inspection of the globular mass.

It was fortunate that the transudate from the strangulated loop of gut was completely localized

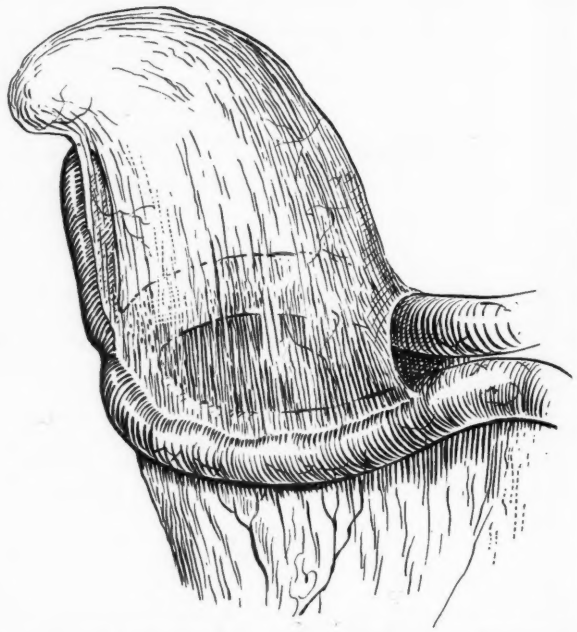

FIG. 492.—Reconstruction of the anatomical configuration prior to the onset of intestinal strangulation.

within the peritoneal sac and was thus able to be released without soiling the general peritoneal cavity. Although the gut was not gangrenous, the obstruction was obviously at a late stage as seen by the proximal distension of the bowel.

Previous References.—Several dozens of references have been examined but it has not been possible to trace a previous example of this nature. Only a small proportion of Meckel's diverticula possess a mesentery, which is usually short and attached to one aspect only, but surprisingly little information can be obtained about its anatomical variations. The remnant of the vitelline artery, itself sometimes a cause of obstruction, was quite prominent in this case but did not play any active part in the mechanism.

SUMMARY

A unique cause of intestinal strangulation has been reported. In a 6-year-old boy an abnormally developed mesentery of a Meckel's diverticulum assumed the form of a peritoneal caul in which the diverticulum itself and the adjacent loop of ileum became incarcerated. Recovery took place after operation.

I am indebted to Mr. A. Philp Mitchell for the opportunity of treating this case, and to Mr. G. Donald for the illustrations.

SOLITARY CÆCAL DIVERTICULITIS

By N. F. KIRKMAN

DICKINSON SURGICAL SCHOLAR, MANCHESTER

SOLITARY cæcal diverticula are distinctly rare, but they form an interesting type of cæcal lesion. As the right lower quadrant of the abdomen is the one most frequently explored, especially by relatively inexperienced operators, cæcal diverticula merit a little more attention than is usually given to them in text-books of surgery. In *British Surgical Practice* (1948, **3**, 273) it is stated briefly that these diverticula are rare and often mistaken for carcinoma and tuberculosis of the cæcum but no details are given.

Rather more than 100 cases have now been recorded (Anderson, 1946). Four cases have been published in British journals during the last twelve years—Bennett-Jones (1937), Grace (1938, 1940), Hendlass (1944), Dorling (1945).

Failure to recognize the condition at operation, owing partly to lack of knowledge of its occurrence, has led to cæcal diverticula being treated by a variety of operations, some of these needlessly extensive and severe (Gatewood, 1945; Jonas, 1940; Busch and Friedfeld, 1942).

It is partly with the hope of preventing such procedures that this article is written.

CASE REPORT

HISTORY.—M. C., female aged 25, married with no children, attended the hospital for admission on Sept. 7, 1948, as a case of acute appendicitis.

FIG. 493.—Showing the cæcum with the diverticulum as seen at operation. The appendix tip is held in Duval's forceps and it is protruding below the ileum. The diverticulum is inflamed.

She had enjoyed good health until two days previously when central abdominal pain began. She had nausea but no vomiting. After 24 hours the pain shifted to the right iliac fossa, but some central umbilical pain remained.

Bowels: Were often constipated, but she had loose motions during the few days prior to admission.

Micturition: Normal. No scalding or frequency.

Menses: Always irregular. L.M.P. two months previously.

ON EXAMINATION.—Temperature 101·1°; Pulse 120; Respiration 22. A well-built woman of average configuration with a flushed face. Tongue coated.

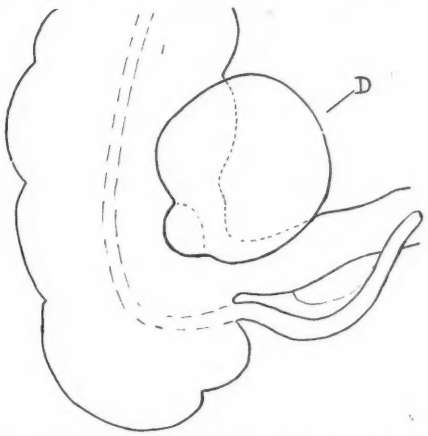

FIG. 494.—Drawing showing the relation of the diverticulum to the cæcum. 'D' indicates diverticulum.

Abdomen: moved well on respiration. There was marked tenderness over the right iliac fossa and on the right flank, with no rigidity. Rectal examination revealed some tenderness in the right side of the pelvis, but no abnormal pelvic masses were detected. Cardiovascular system and respiratory system were normal. B.P. 125/90. A pre-operative diagnosis of acute appendicitis was made, with the probability of the appendix having perforated and given rise to local peritonitis.

AT OPERATION (Sept. 7).—Under general anæsthesia, nitrous oxide, oxygen, and ether, an oblique muscle-cutting approach (Rutherford Morison, Grey Turner 1943) was made to the right iliac fossa. The appendix was displayed in the right iliac fossa, with its tip at the pelvic brim; it was found to be normal. The cæcum, which was mobile, but not abnormally so, was examined and it was found that on the inner side in the ileo-cæcal angle there was a firm rounded yellowish-pink swelling approximately 2 in. (5 cm.) in diameter attached to the cæcum. An examination of the adjacent mesentery revealed no lymphatic glandular enlargement and the condition was considered to be a simple tumour of the cæcum with some inflammation around it. *Figs.* 493, 494 show the condition which confronted the operator. As the lesion occupied the ileocæcal angle local removal was deemed impracticable, so the cæcum together with 3 in. of terminal ileum was resected by a Paul-Mikulicz procedure. The cut ends of the bowel were both exteriorized. A colotomy tube was inserted into the lower end of the ileum, and the proximal end of the ascending colon was held adjacent to the ileum in the wound by Schoemaker forceps.

POST-OPERATIVE PROGRESS.—Glucose-saline was given by intravenous drip for 24 hours, and sulphamezathine 1·0 g. was administered 4-hourly.

Sept. 11: The pyrexia had subsided. The ileostomy was draining well and the general condition was good.

Sept. 21 : An enterotome was applied to crush the spur between the bowel ends.

Oct. 4 : The enterotome was removed, the patient was getting up.

Oct. 9 : Œdema of the ileostomy due to crushing of the spur was subsiding and a bowel action per rectum occurred.

Nov. 8 : The ileostomy was closed by operation.

Nov. 22 : The wound had healed, the bowel action was normal, and the patient was discharged from hospital quite fit.

DISCUSSION

Other cæcal conditions such as carcinoma, tuberculosis, actinomycosis, Crohn's disease, and simple ulcer must be distinguished from diverticulitis of the cæcum.

The cardinal features of diverticulitis which aid differentiation are : a history very suggestive of acute appendicitis in a patient often under 40 years of age, and palpation of the mass through the opposite cæcal wall which reveals the mouth of the diverticulum (Moon and Schenk, 1945 ; Kennon, 1933).

Carcinoma of the cæcum is usually a fungating tumour ; the ulcerating type is rare and should be differentiated from a diverticulum by its firmness and the shallow irregular deficiency in the cæcal wall which is unlike the circular symmetrical opening of a diverticulum.

Tuberculosis of the cæcum is a more chronic condition than diverticulitis, small tubercles are usually found on the peritoneum, and the condition is less definitely limited to a solitary circumscribed mass. There is marked ileocæcal and mesenteric adenitis, which is often widespread and gross. Actinomycosis is characterized by chronicity and massive fibrosis, with usually a surprisingly small quantity of pus present in multiple sinuses.

In Crohn's disease the condition is rarely so acute, there is usually generalized thickening of the adjacent ileum, the ileum being as a rule more affected than the cæcum, and marked mesenteric adenitis is usually present.

COMMENTARY

Symptomatology.—The chief signs and symptoms of solitary cæcal diverticulitis—central abdominal pain moving to the right iliac fossa, nausea and vomiting, with right-sided abdominal tenderness—are identical with those of acute appendicitis, as one would expect them to be. In most cases reported the pre-operative diagnosis has been acute appendicitis. Schnug (1943) gives the incidence of vomiting as only 26 per cent of cases in cæcal diverticulitis as against 73 per cent of cases in acute appendicitis. This difference, however, is hardly sufficient to aid in differentiation, as mentioned by Baker and Carlisle (1943).

Operative Diagnosis.—Approximately two-thirds of the recorded cases have been correctly diagnosed as cæcal diverticulitis at operation (Schnug, 1943). Most of the remaining one-third not recognized at operation were treated by a colectomy, often a complete right-sided hemicolectomy.

When seen at operation the diverticulum may be inflamed, thickened, indurated, or contain solid fæces—as in the case recorded above. These conditions render diagnosis more difficult.

Treatment.—The treatment required is usually merely the removal of the diverticulum (Johnson, 1944). In most cases the operator, recognizing the condition, should have no difficulty in excising the diverticulum, or a wedge resection of the adjacent cæcum may be performed if necessary (Dorling, 1945). Diverticula have been successfully treated by marsupialization (Frehling, 1945) and by inversion (Hendlass, 1944). Schnug (1943) points out that the drainage of the inflamed diverticulum into the bowel is good, and he suggests that the diverticulum may be left in situ with external drainage of the inflammatory mass.

The point to remember is that having recognized the diverticulum at operation no major colectomy should be done, but merely the simplest eradication of the condition. In the case reported the position of the diverticulum and its inflammatory state rendered excision somewhat hazardous and a local resection of the cæcum was carried out. This seems to be the most extensive operation that is likely to be needed.

Aetiology.—The cause of these diverticula is still in doubt. They are probably not closely related to the diverticula which occur in the distal parts of the colon. The latter occur chiefly in men over 50 years who are fat and constipated (De Witt and Abeloff, 1938). Anderson (1946) points out that 6 per cent of patients who have had the colon X-rayed have had diverticulosis present in the distal parts. The solitary cæcal diverticulum, however, occurs chiefly below the age of 40 and equally amongst the sexes.

Some authors (Dorling, 1945 ; and Schnug, 1943) have classified the solitary cæcal diverticulum into true or false according to the presence or absence of all the bowel coats in the walls of the diverticulum. Unless key-block serial sections of the whole diverticulum are cut it is difficult, if not impossible, to be certain that these diverticula have complete layers in their walls. In many recorded cases no differentiation between a true or a false diverticulum is made (Jonas, 1940). Some of them are obviously false, arising from traction on the cæcal wall by adhesions to neighbouring structures (Dorling, 1945 ; and Grace, 1940). Others appear to be truly congenital. Diverticula have been recorded as arising from all aspects of the cæcum. It has been postulated that the true congenital cæcal diverticulum is a misplaced Meckel's diverticulum, but a case has been recorded where a Meckel's diverticulum and a true solitary diverticulum were both present in a girl aged 3 years (Odquist and Petren, 1931). These authors suggest that the diverticulum arose in embryonic life from a totipotent cell in a cæcal haustration. This seems a reasonable but not a completely convincing solution to the problem of the origin of the true solitary cæcal diverticulum.

SUMMARY

1. A case of solitary diverticulitis of the cæcum is recorded.

2. It is stressed that the simplest surgical measures only are required for treatment, e.g., diverticulectomy.

3. The possible origins of these diverticula are discussed.

I wish to thank Mr. H. T. Cox for permission to operate on and treat this case; Dr. Stent for pathological investigations; and Professor Wood Jones for help in preparing this paper.

REFERENCES

ANDERSON, L. (1946), *Proc. Mayo Clin.*, **21**, 465.
BAKER, J. W., and CARLISLE, T. (1934), *J. Amer. med. Ass.*, **122**, 354.
BENNETT-JONES, M. S. (1937), *Brit. J. Surg.*, 25, 66.
BUSCH, I., and FRIEDFELD, L. (1942), *Amer. J. Surg.*, **57**, 555.
DORLING, G. C. (1945), *Brit. med. J.*, **1**, 513.
FREHLING, S. (1945), *Ann. Surg.*, **121**, 83.
GATEWOOD, J. W. (1945), *Ibid.*, **122**, 52.
GRACE, A. J. (1938), *Brit. J. Surg.*, **26**, 200.
— — (1940), *Surgery*, **7**, 232.
HENDLASS, R. F. (1944), *Brit. med. J.*, **2**, 309.
JOHNSON, T. A. (1944), *Gastro-enterology* (Bockus), **2**, 705. Philadelphia and London: W. B. Saunders.
JONAS, A. (1940), *J. Amer. med. Ass.*, **115**, 194.
KENNON, R. (1933), *Brit. J. Surg.*, **20**, 521.
MOON, Z. B., and SCHENK, H. L. (1945), *Amer. J. Surg.*, **68**, 364.
ODQUIST, B., and PETREN, T. (1931), *Virchows Arch.*, **280**, 580.
SCHNUG, E. (1943), *Surgery*, **13**, 282.
TURNER, G. GREY (1943), *Modern Operative Surgery*, 3rd. ed. London: Cassell.
DE WITT, S., and ABELOFF, A. J. (1938), *J. Mt. Sinai Hosp.*, **4**, 781.

PERISEMILUNAR DISLOCATION OF THE CARPUS

A CASE OF PERISEMILUNAR-TRANSTRIQUETRAL DISLOCATION

BY J. N. WILSON
ORTHOPÆDIC REGISTRAR, COVENTRY AND WARWICKSHIRE HOSPITAL

POSTERIOR dislocation of the carpus around the semilunar bone, with normal position of the latter in relationship to the radius, is one of the rarer types of injury to the wrist. The same injury associated with fracture of the scaphoid bone is also recognized as a rare condition. The case described below is not mentioned in the majority of descriptions and must therefore be a very rare combination of fracture and dislocation.

CASE REPORT

J. H., a young man of 23, was seen in the Casualty Department on Jan. 22, 1949, following an injury to the right wrist. He had been starting the engine of a lorry when the handle kicked back on him, forcibly dorsiflexing the wrist. He felt immediate severe pain in the front of the wrist and noticed that a swelling had appeared at the part where he felt the pain and that this swelling was pulsating. He was seen by the House Surgeon on duty, who observed that there was considerable tenseness under the skin over the anterior carpal creases and that the radial pulse was hardly perceptible. The wrist was X-rayed but was reported as being normal.

The joint was therefore immobilized in a plaster-of-Paris back slab and the patient asked to attend the following day.

Twenty-four hours later the patient was seen again. By this time he was in considerable pain and was complaining of tingling of the fingers. The whole hand was swollen and the anterior surface of the wrist was tense and there was pulsation over the swelling on the radial side, which later proved to be transmitted pulsation from the radial artery. Clinically there was definite 'dinner

FIG. 495.—Showing perisemilunar dislocation with possible fracture of the triquetrum.

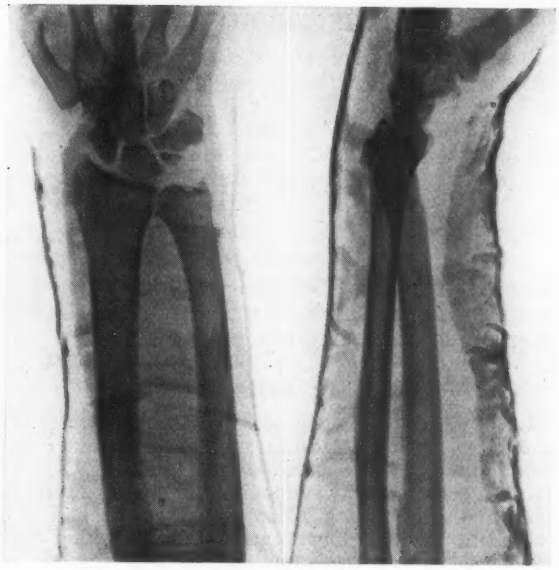

FIG. 496.—Shows reduction of dislocation and fracture of the triquetrum.

fork' deformity and when the radiograph was again scrutinized it became obvious that the patient was suffering from a posterior perisemilunar dislocation. It was noticed that in the anteroposterior view there appeared to be some deformity of the triquetrum. Therefore a provisional diagnosis of perisemilunar-transtriquetral dislocation was made (*Fig.* 495).

Reduction was carried out fairly easily by traction, dorsiflexion, and then palmar flexion, and the limb was immobilized in a short plaster cast. After reduction the patient's symptoms were completely relieved and the swelling of his fingers rapidly subsided. Post-reduction X-ray examination (*Fig.* 496) showed that the dislocation had been completely reduced and that the provisional diagnosis of concurrent fracture of the triquetrum had been correct. Progress was uneventful. The wrist was immobilized for six weeks and despite the fact that the triquetrum did not look very firmly united (*Fig.* 497)

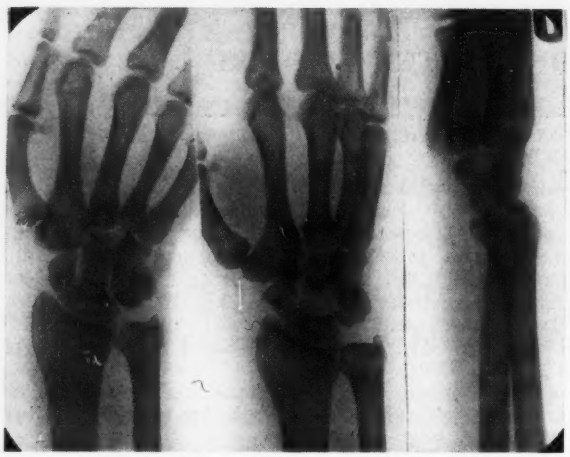

FIG. 497.—Radiograph taken six weeks after reduction.

the clinical result was excellent and the patient was able to start work just two months after the injury.

DISCUSSION

It is generally accepted that fracture-dislocations of the wrist result from dorsiflexion injuries, the usual history being that of a fall on to the outstretched hand. MacAusland (1944) describes the mechanism of all semilunar dislocations as one of preliminary perisemilunar dislocation with a possible further stage of complete isolated dislocation of the semilunar bone. He states that this may or may not be associated with fractures of the other carpal bones, but gives no explanation for the variance. It seems possible that the associated fractures may be due to the degree of rotation which occurs at the time of injury. If this is so the elongated oblique shape of the scaphoid renders it more likely to injury, especially when a fall on the hand is more likely to have its main rotatory force centred on the base of the thenar eminence. By virtue of its small size and different shape the triquetrum is much less vulnerable in such an injury. However, it is possible that in the case described above the precipitating force was centred over the hypothenar eminence by virtue of the fact that the end of the starting handle would be held in relation to this part of the hand. It is interesting to note that there was no evidence of the commonly associated fracture of the styloid process in this case. Whether this is of any aetiological significance it is difficult to say.

Kellam and McGoey (1945) record 2 cases of retrolunar triquetrum dislocation which it has been assured were of the same nature as the present case. Unfortunately, however, there is no description of the individual cases nor any X-ray photographs for comparison. Their figures, however, show the injury to be just over 1 per cent of the total number of cases of carpal injuries reviewed and 22 per cent of the total number of fracture-dislocations of the wrist. These figures help to underline the relative rarity of this particular type of injury.

REFERENCES

KELLAM, H. I. J., and MCGOEY, P. F. (1945), *Canad. med. Ass. J.*, **53**, 332.
MCAUSLAND, W. R. (1944), *Surg. Gynec. Obstet.*, **79**, 256.

A CASE OF 'IDIOPATHIC' GANGRENE OF THE SCROTUM

BY D. H. RANDALL, SHEFFIELD

IDIOPATHIC gangrene of the scrotum is not an excessively rare condition, but is of sufficient interest to merit each case being reported.

CASE REPORT

HISTORY.—A previously healthy man of 36 years was admitted to the Royal Infirmary, Sheffield, under Mr. Clifford Jones on the afternoon of Monday, Oct. 18, 1948, in a severely toxic state and obviously critically ill.

Up to the preceding Friday, Oct. 15, he had been in excellent health. That evening on returning from work he had felt ill, was sweating, and had a severe dull pain in his head, shoulders, and back; he had diarrhœa and anorexia. He was seen by his doctor, who treated him for a 'a chill'. The next day he was quite well. At 8 p.m., two days later, Oct. 17, whilst sitting by the fire he noticed his scrotum was swelling as it felt tight within a suspensory bandage he normally wore. There was a gradual onset of burning pain in the scrotum which became intolerable, and he sent for his doctor, who observed a moderate swelling and slight redness of the whole scrotum but nothing else. The next morning he felt worse, the pain was more severe and the scrotum was more swollen, with purplish patches on it. This was fourteen hours after onset of the pain and swelling. He was admitted to the Royal Infirmary.

Throughout there had been no urinary disturbance whatsoever. There was no history of trauma of any kind.

ON ADMISSION.—The patient was obviously in great pain and desperately ill. He had a rigor immediately after admission. Temperature 104°, pulse 126.

The scrotum was swollen, measuring 8 in. across, with a purplish-black patch of superficial gangrene over the whole anterior surface. It was tender and œdematous. The black areas were quite cold and leathery, whilst the areas of normal skin were pinkish red and hot to the

touch. The condition was limited to the scrotum, there being no other visible abnormality, but there was tenderness over both inguinal canals. There was no evidence of any abrasions or septic spots, etc., on the scrotum. The liver was just palpable and tender.

Rectal examination was negative.

The diagnosis made was a fulminating gangrene of the scrotum, probably infective in origin. In appearance

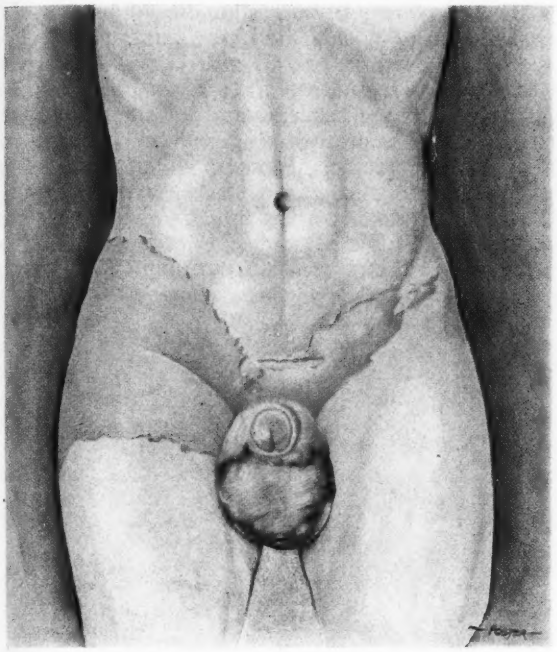

FIG. 498.—Drawing made on Oct. 21, 1948. Showing extent of gangrene and manner of spread of the infection along the tissue planes, the right side being wider than the left.

the scrotum and lower abdominal wall resembled an extravasation of urine, but there were no urinary symptoms except albuminuria. There was a slight polymorpholeucocytosis.

He was treated with large doses of penicillin, sulphamezathine, and forced fluids.

The next morning his general appearance had improved—the temperature was 101°, the pulse 120, and the pain had gone. The scrotum was a little less swollen but redness was now apparent in both groins. On the right side it extended as far as the anterior superior iliac spine and spread about 1 in. above and below the inguinal ligament, whilst on the left it was confined exclusively to the region of the inguinal canal. The skin was hot and the margin of the inflamed area definitely raised.

The general condition remained good, temperature 99°, pulse 90.

The whole of the anterior surface and the lower third of the posterior surface of the scrotum was definitely gangrenous, with a sharp line of demarcation. The area of redness extended out into the loins and well up on to the abdominal wall. There was minute vesiculation (*Fig.* 498). Swabs taken from the gangrenous scrotum showed an aerobic culture, *B. proteus*, and *Ps. pyogenes*. *Anaerobic culture revealed a heavy growth of hæmolytic streptococcus.*

The subsequent progress of the case was uneventful. He had altogether 32 g. of sulphamezathine and 24 mega units of penicillin. The gangrenous areas of the scrotum separated and healed rapidly. No skin-grafting was necessary.

Comment.—The interesting features of this particular case were :—

1. Its occurrence in a relatively young (36 years old), fit man.

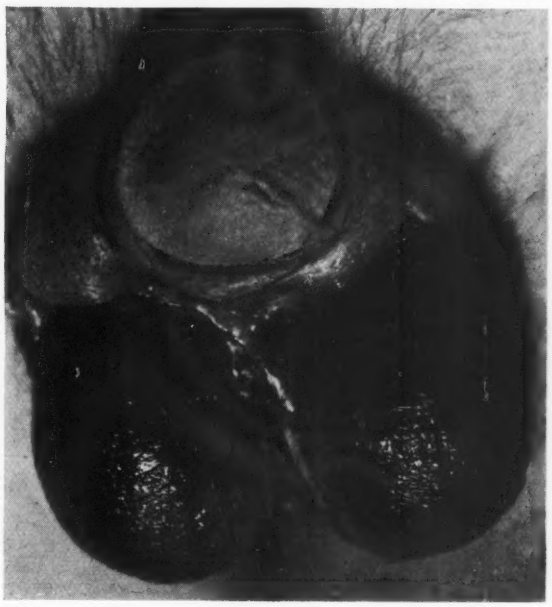

FIG. 499.—Photograph showing the state of affairs after the sloughs had separated.

2. Complete absence of any obvious predisposing factor or portal of entry for the organisms.

3. The amazingly rapid onset of the gangrene —the skin was purplish black and cold within 14 hours of the first local symptoms.

4. The absence of local signs of severe infection prior to the onset of the gangrene.

5. The spread of the infection along the plane deep to Colles' fascia as in extravasation of urine from the bulbous urethra.

6. Complete absence of any urinary symptoms throughout the course of the disease.

7. The dramatic response to penicillin and sulphamezathine.

My thanks are due to Professor R. St. Leger Brockman and Mr. Clifford Jones for permission to publish this case.

BIBLIOGRAPHY

MAIR, G. B. (1945), *Lancet*, **1**, 464.
MANSFIELD, O. T. (1946), *Brit. J. Surg.*, **33**, 275.

A RECURRENT BENIGN GIANT-CELL SYNOVIOMA OF TENDON-SHEATH OF 34 YEARS' DURATION

By M. J. STEWART and C. J. E. WRIGHT

FROM THE DEPARTMENT OF PATHOLOGY AND BACTERIOLOGY, UNIVERSITY OF LEEDS

In 1915 Stewart and Flint recorded 2 cases of benign giant-cell synovioma of tendon-sheaths. One of these—their second case—was that of a woman of 27, first seen in 1911, who complained of a swelling on the front of the left wrist. This had appeared about two and a half years before, soon after a fall in which she had injured her wrist. The swelling had been almost continuously painful and by the time she sought medical relief had attained the size of a walnut. A typical myeloid tumour of the tendon-sheath (benign giant-cell synovioma) was removed in October, 1911. There had been no sign of recurrence when the patient was last visited in January, 1915.

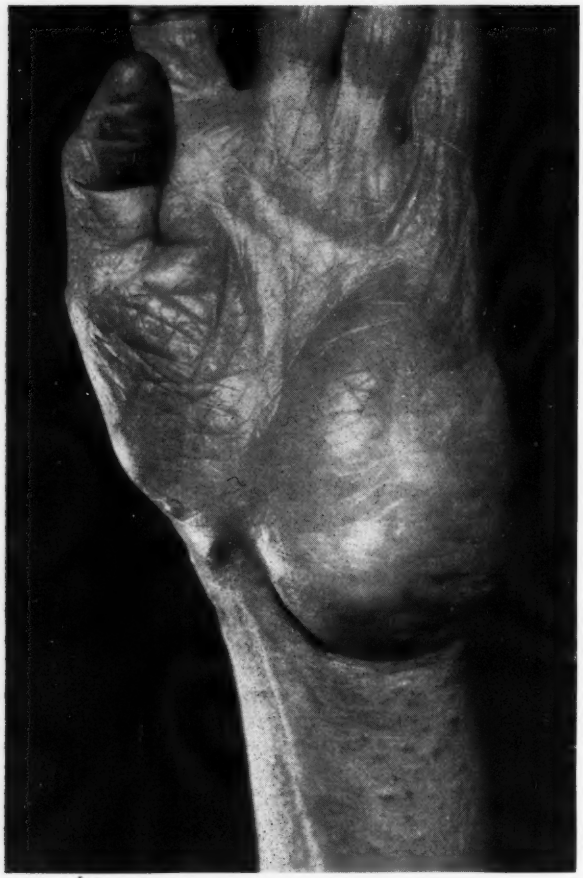

FIG. 500.—Recurrent benign giant-cell synovioma of a flexor tendon-sheath. The primary tumour was removed thirty-seven years ago, and it is thirty-four years since the recurrence was first noticed. Its rate of growth has been uniformly slow throughout this period.

In the course of a systematic follow-up of a series of cases of this kind, the patient was again visited in November, 1948, nearly thirty-four years later, and was found to have a recurrence. This had made its appearance soon after she was seen in 1915, and had continued to grow, very slowly, ever since. The recurrent tumour is situated on the flexor aspect of the left wrist and extends on to the palm of the hand (*Fig.* 500). It measures about 3 in. in length by 2½ in. in breadth and is raised about 1 in. above the surrounding skin surface. The tumour is soft, especially around the periphery, and there is a faint linear depressed surgical scar 2 in. in length in the long axis of the tumour, to which it is apparently attached. The patient is in good health and has a perfectly useful hand, her only complaint being of occasional stabbing pain and the unsightliness of the tumour, but as these do not trouble her unduly she will not consider a further operation.

COMMENTARY

In their original report, Stewart and Flint expressed the opinion that, as the benign giant-cell synovioma of tendon-sheaths is to all intents and purposes a benign growth, conservative treatment should be adopted. "The tumour should be excised locally; it can usually be shelled out with considerable ease; and in the event of recurrence, we think that a second local operation should be given a trial before anything radical is done." Cases of recurrence have indeed been reported on a number of occasions, but we see no reason for suggesting any departure from this practice unless the original tumour is of rapid growth and histologically anaplastic. It is perhaps rather surprising that recurrence is not more frequent, since this particular variety of tendon-sheath tumour is often much lobulated, and it would be easy to leave a small portion behind in excising the growth.

In a review of 156 cases where the information was available (Galloway, Broders, and Ghormley, 1940), the average duration of the primary tumour at the time the patient was first seen was five years and the longest thirty-one years. We have been unable to discover any record of a recurrent tumour of such long duration as the one now reported.

REFERENCES

GALLOWAY, J. D. B., BRODERS, A. C., and GHORMLEY, R. K. (1940), *Arch. Surg*, **40**, 485.

STEWART, M. J., and FLINT, E. R. (1915–16), *Brit. J. Surg.*, **3**, 90.

A CASE OF DIRECT INGUINAL HERNIA ILLUSTRATING IMPROVEMENT IN TONE OF THE OBLIQUE MUSCULATURE OF THE ABDOMINAL WALL AS A RESULT OF SURGICAL REPAIR OF THE HERNIA

PRELIMINARY REPORT

By DAVID H. PATEY
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THE common type of direct inguinal hernia is a diffuse bulge of the posterior inguinal canal with associated stretching of the transversalis fascia, and the essential principle of all the different techniques of surgical treatment is the repair and strengthening of the stretched area. One of the standard methods is that of the ligamentous darn with fascia lata sutures, and in a previous communication (Patey, 1949) it has been shown that the direction of insertion of the sutures is of practical importance, in that better support is given to the weakened area if the sutures are inserted from below upwards and outwards so that they are tautened by the contraction of the internal oblique. The case herewith reported, in which this method of repair was adopted, would seem to show that more is involved than the mere local strengthening of a weakened area, and that the repair of the hernia may also have a beneficial effect on the whole of the oblique musculature of the corresponding side of the abdominal wall.

FIG. 501.—Before operation, showing the bulging of the oblique regions on either side as the intra-abdominal pressure was increased by raising the shoulders.

CASE REPORT

The patient was a man, aged 44, with a right inguinal hernia of two months' duration. He was a thin type, and the abdominal postural tone was poor, so that on standing the abdomen was protuberant and sagging. When asked actively to brace up his abdominal muscles, however, the abdomen became scaphoid in shape. The deficiency in postural tone was more marked in the obliques than in the recti, as could be readily demonstrated by a few simple tests. Thus when he was asked to give an expiratory strain, the lateral parts of the abdominal wall bulged. A convenient way to demonstrate these lateral bulges was to let the patient lie relaxed on his back on a couch, and then to get him slowly to raise his shoulders. The recti came into action with the beginning of the movement, but the oblique regions of the abdomen remained relatively flaccid so that they bulged with the increased intra-abdominal pressure. (*Fig.* 501). It was only when the shoulders had been raised a considerable way that the oblique muscles came fully into action and the bulges disappeared. This delay in synergic action of the oblique musculature under

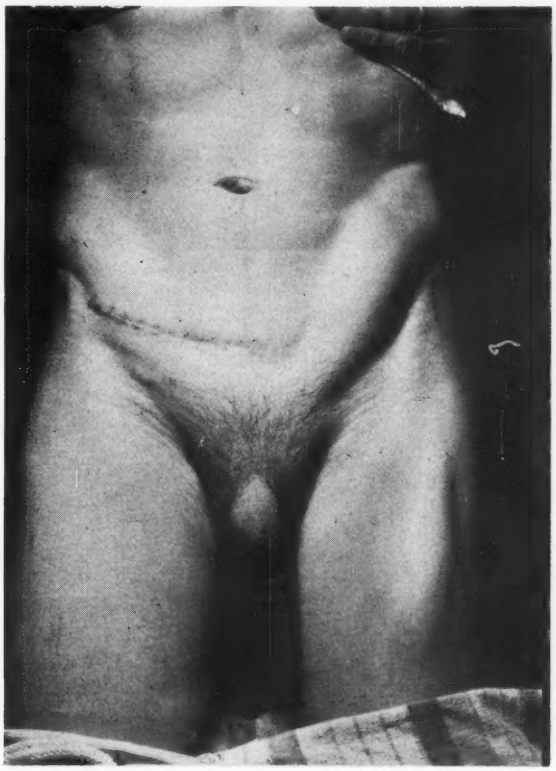

FIG. 502.—After operation on the right inguinal hernia, showing the diminished bulging of the oblique region on the operated side.

conditions of strain presumably diminished the protection normally given to the inguinal region and predisposed to the inguinal hernia.

A subsidiary indication of the deficient action of the oblique muscles was furnished by the behaviour of the cremasteric muscles. When the patient stood and braced up his abdominal muscles, the testicles were pulled up at the same time. If, however, he gave an expiratory strain so that the oblique regions bulged, the cremasteric muscles did not come into action and the testicles were not pulled up.

The patient was admitted to hospital, and the operation was delayed for ten days to see if an intensive course of exercises would improve the postural tone of the abdominal muscles. No change was apparent. Operation was therefore performed, and a direct inguinal hernia of the diffuse bulge type found. The stretched transversalis fascia was plicated, and the weak area between the upturned edge of the inguinal ligament and the conjoint muscle and tendon strengthened by a ligamentous darn suture of fascia lata. The suture was inserted so as to run obliquely from below upwards and outwards as described above, and under minimal tension. The external oblique was sutured superficial to the cord by a running fascia lata suture.

The post-operative course was uneventful and the wounds healed by first intention. When tests were performed on the abdominal muscles after operation, there was seen to be a marked difference in the tone and synergic action of the oblique muscles on the operated and non-operated sides. Thus when the patient contracted the abdominal muscles by raising his shoulders from the bed as before operation, the oblique bulge on the operated side was much less marked and disappeared much sooner during the course of the movement than on the non-operated side (*Fig.* 502). In other words, the tone and synergic action of the whole of the oblique musculature on the operated side was much improved by the operation. Injection of novocain into the edge of the internal oblique muscle corresponding to the level of insertion of the fascia lata sutures did not effect this improved tone, which has remained well marked up to the present time—three months after operation. No conclusions were possible on the effect of the hernial repair on cremasteric action in this case as the function of the cremaster on the operated side was almost destroyed by the operation.

DISCUSSION

The operation on the direct inguinal hernia in this case not only repaired the hernial area but resulted in an improvement in the tone of the whole oblique musculature on the operated side. The mechanism of production of the improved tone was presumably by the stretch reflex, brought into action by the pull of the new fascia lata ligaments on the internal oblique as the intra-abdominal pressure was raised. Such being the case, the question of the best method of insertion of fascia lata sutures in a hernia repair is not merely a problem of local repair, but one which involves also the question of the most efficient way of stimulating the stretch reflex. From the latter point of view the direction of insertion of the sutures becomes of particular importance, and it would seem that an oblique upward and outward direction of the sutures not only affords an efficient protection to the hernial area but is calculated also to excite the stretch reflex. The question of the tension at which the sutures are inserted also becomes of importance, and the optimum tension can be defined as that tension which best excites the stretch reflex without cutting through the tissues into which the suture is inserted.

If the stretch reflex brought into action by the repair of a weakened transversalis fascia is of importance, it is probable that the same mechanism brought into action by the healthy transversalis fascia is also of importance. Some surgeons have had difficulty in the past in accepting such a thin and relatively weak membrane as the transversalis fascia as playing the part in the problem of hernia that has been claimed for it. If, however, the transversalis fascia is not merely a passive barrier but also a trigger mechanism for the stretch reflex, some of these difficulties are resolved.

Finally, surgical experience on the survival of fascia lata sutures in hernia repair is twofold. Sometimes subsequent operation reveals the fascial sutures clearly recognizable as such; at other times they seem to disappear completely. It is tempting to speculate on whether or not this depends on the new ligaments formed by the fascial sutures being put to functional use. If it does, the insertion of the sutures in such a manner as to ensure their functional use becomes of importance from the point of view of graft survival. It will be interesting to note, too, whether the improved tone on the operated side is maintained, or whether the stretch reflex brought into action by the fascial sutures is gradually lost just as was that of the original transversalis fascia.

SUMMARY

1. A case of direct inguinal hernia with associated deficient tone and synergic action of the oblique musculature of the abdominal wall is described.

2. Fascial repair of the hernia resulted also in a marked improvement in this deficient tone and synergic action.

REFERENCE

PATEY, D. H. (1949), *Brit. J. Surg.*, **36**, 264.

REVIEWS AND NOTICES OF BOOKS

Lehrbuch der Chirurgie (Garrè-Stich-Bauer). Revised by Dr. RUDOLF STICH and Dr. KARL-HEINRICH BAUER. Fourteenth and fifteenth edition. 6½ × 9¾ in. Pp. 860 + xii, with 601 illustrations, some coloured. 1949. Berlin: Springer. DM. 60.

THIS *Lehrbuch der Chirurgie* was first published in 1920 by Professors C. Garrè and A. Borchard. That the edition before us is the fourteenth/fifteenth may be accepted as evidence of its popularity and representative character as a text-book. Professors Stich and Bauer, of Göttingen and Heidelberg respectively, have undertaken this latest revision. They state that difficulties, both personal and general, have delayed its appearance and account for a five-year gap since the thirteenth edition.

Garrè's scholarly if somewhat sententious introduction entitled The Science and Study (*Lehren und Lernen*) of Surgery is retained. The attempt to compress the whole subject into one volume has been continued. Few of the classical illustrations have been discarded, but a good number of new and instructive photographs have been introduced.

An indication of the comprehensive character of the work may be given by recording the presence of a seven-page section on anæsthesia (general and local) and ten pages devoted to diseases of the teeth, which include precise instructions and speaking figures of methods of extraction. The book concludes with a fully illustrated account of the formal ligature of the main arteries, together with a section on amputations. In the latter it is noticeable that but slight attention is drawn to the level of bone section and its relation to the nutrition and utility of the stump. The fact that descriptions and illustrations of Lisfranc, Chopart, and Pirogoff amputations are retained suggest that this chapter is designed to assist the examiner rather than the practical surgeon.

Under the ruling circumstances it is not surprising that the treatment of wound infection should appear somewhat out of date to the western surgeon. The merits and uses of many chemical antiseptics are considered in detail and the impression is given that the antibiotic agents have not yet found a firm place in the authors' practice. This is especially noticeable in reference to the management of acute osteomyelitis—early and radical operative procedures are advocated as of the first importance.

The division of the contents is on a topographical basis and various injuries are considered under appropriate headings. The subject of trauma of the head and extremities is given liberal space.

A student's text-book should give a clear and sound exposition of principles and is not expected to chronicle all the latest advances or changes of fashion in the technique of the subject. Although this work fully conforms to the primary demand, it must be confessed that it goes beyond reasonable conservatism in so far as it omits reference to many novel procedures which have been developed in America and this country during the last five or ten years. The isolation of Germany from western influence is fully exposed when one finds little or no reference to what seems to us well-established advances. In the realm of thoracic surgery radical operation for carcinoma of the bronchus is hardly considered, no mention is made of thymectomy or the arterial surgery of cardiac malformations, yet considerable space is given to Trendelenberg's operation for the removal of a pulmonary embolus. The symptomatology and surgery of the prolapsed intervertebral disk (*Bandscheibenvorfall*) is viewed very briefly and cautiously.

The chapter on abdominal hernia is an outstanding example of full and clear presentation of the pathological anatomy and clinical manifestations of the various conditions. The illustrative photographs are admirable. The operative treatment of inguinal hernia is practically confined to a description of the Bassini operation. The recurrences from this procedure are recorded as being between 0·5 and 3 per cent—few British surgeons would claim comparable results for this operation. It is of interest to note that throughout the authors have little faith in radiotherapy in the treatment of malignant disease and whenever possible advise radical surgery.

Despite the above limitations the work continues in a limited compass a remarkably complete review of general surgery in the widest sense. It is inspired throughout with a sound conservative approach to principles and sets a high ideal in surgical conduct.

The volume is well bound and printed on good paper. The 600 figures come out well and mostly serve a useful purpose.

X-ray Diagnosis. For Clinical Students and Practitioners. By G. SIMON, M.D., B.Ch. (Cantab.), F.F.R., Assistant Radiologist at St. Bartholomew's Hospital and Assistant Director of the X-ray Department, The Brompton Hospital. 9½ × 7¼ in. Pp. 208 + x, with 180 illustrations. 1949. Cambridge: W. Heffer & Sons Ltd. 20s. net.

THIS is a book which the clinician has been waiting for and its publication is long overdue. It is a book which the medical student and hospital resident medical officer will find of great value, for it gives them the information they require in the interpretation of X-ray films.

Commencing with an introductory section explaining the various X-ray techniques and the preparation of the patient for the various examinations, the author has arranged his subject-matter on a clinical rather than on an anatomical basis.

The part concerned with radiology in bone and joint diseases is strikingly good and the radiographs are excellent and beautifully reproduced. The reader obtains a very good idea of the various X-ray appearances in different bone and joint diseases.

The section devoted to radiology of the abdomen is very practical, and here again the radiographs are excellent. Perhaps the chapters devoted to radiology of the chest are the best in the book, but this is not to be wondered at because the author is the Assistant Director of the X-ray Department of the Brompton Hospital.

This book will take its place at once as one of the really useful books for the student and practitioner.

Essentials of Orthopædics. By PHILIP WILES, M.S., F.R.C.S., Orthopædic Surgeon, Middlesex Hospital. Large roy. 8vo. Pp. 486 + xv, with 7 colour plates and 365 text-figures. 1949. London: J. & A. Churchill Ltd. 42s.

THIS book covers the essentials of orthopædics very well; it is written in a clear, concise, and dogmatic manner, and this is what is wanted in a text-book to-day. Orthopædic surgery is a large and ever-growing branch of surgery, and Mr. Wiles has managed to cover this large subject in a complete and masterly fashion and has illustrated his book with first-class coloured drawings, clinical photographs, and radiographs.

One would have liked to have seen more about the surgery of the hand, especially the treatment of infection of the tendon-sheaths. This is what the student and practitioner require, and it is far more important than

tumours of the tendon-sheaths. However, there is little to criticize in this very readable and practical book. It covers the essentials of orthopædics in an admirable way.

British Surgical Practice. General Editors: Sir ERNEST ROCK CARLING, F.R.C.S., F.R.C.P., Consulting Surgeon, Westminster Hospital; and Sir JAMES PATERSON ROSS, K.C.V.O., M.S., F.R.C.S., Surgeon and Director of Surgical Clinical Unit, St. Bartholomew's Hospital; Professor of Surgery, University of London. In eight volumes. $6\frac{3}{8} \times 8\frac{3}{4}$ in. Volume VI. Pp. 554 + xxvi, with Index, 303 illustrations, 4 plates. 1949. London: Butterworth & Co. Ltd. Price per volume £3, Index £1.

THE sixth volume of this comprehensive work contains some thirty-seven articles on a wide variety of surgical conditions from malingering to peritonitis. The contributors are all men of experience, with a wide knowledge of their subject. It is difficult to single out any article for especial praise because the whole volume is uniformly good. The illustrations are well chosen, many of them are original and are beautifully reproduced.

We look forward to seeing the last two volumes. The work without doubt is the best that has so far appeared in the English language.

Lord Lister. His Life and Doctrine. By DOUGLAS GUTHRIE. $6\frac{1}{4} \times 8\frac{3}{4}$ in. Pp. 128 + viii, with coloured frontispiece and 20 plates. 1949. Edinburgh: E. & S. Livingstone. 15s.

ALTHOUGH there are several books devoted to the life of Joseph Lister yet there is always room for another, especially in the present generation who knew him not, and do not fully appreciate the value of his contribution to surgery and mankind.

Professor Guthrie, as a medical historian, has added to our knowledge of Listeriana in writing this book, for he has selected some interesting pictures and autograph letters which have not been published before. It is curious to note that Lister in 1878 discussed with a former student the study of bacteria and moulds, including penicillium.

This book is written in a stimulating and lucid style and gives the reader a very good mental picture of Lister's life and doctrine. It will appeal both to the undergraduate and graduate.

The Portraiture of William Harvey. By GEOFFREY KEYNES, M.A., M.D., F.R.C.S. Emeritus Surgeon to St. Bartholomew's Hospital. $7\frac{3}{8} \times 9\frac{7}{8}$. Pp. 42, with 32 pp. of plates bearing 43 illustrations. 1949. London: Royal College of Surgeons of England. 25s.

IN 1912, the Council of the Royal Society of Medicine founded, with Sir William Osler as its true parent and first President, a separate Section devoted to the study of the history of medicine. Such a project had been proposed more than once before Osler's coming to Oxford, but had invariably fallen through; it required all his enthusiasm and quickening influence to bring the plan to full fruition. As the first fruit of the new Section's activity there appeared in 1913 from the Oxford University Press a handsome fasciculus entitled *Portraits of Dr. William Harvey*; it contained reproductions in collotype of the then known portraits of Harvey, some sixteen in number, to which were added a *catalogue raisonné* of some two score engravings and an account of several busts and medals struck in his likeness. The publication proved that contemporary portraits of Harvey were more numerous than had been expected, but that there were also many portraits of elderly seventeenth-century gentlemen with peaked beards and white collars which, although labelled with his name, were spurious.

Last year Mr. Geoffrey Keynes—*vir urbanus et instructus*—selected as the subject for his Vicary Lecture at the R.C.S. a more up-to-date review of the Harvey portraits. Which of the many were reliable and authentic? Vicary and Harvey had each in his day served the lecturer's own hospital, St. Bartholomew's, with singular fidelity, and their names are written indelibly on Britain's roll of medical fame. Hero-worship Mr. Keynes admits to be a common human weakness: we all like to know what our heroes looked like while they lived and strove. The impulse to know details of the great man's physical appearance is both widespread and generous, and of the many great figures of the past none should capture the medical man's imagination more readily than that of William Harvey. To this factor Mr. Keynes attributes the surprising number of the portraits which he has found in his search. Demand has increased the supply, and the richness of the supply has added considerably to the difficulties of the honest iconographer. He has examined, with an informed and critical eye, no fewer than thirty-four alleged portraits of the Master, whether executed in oils, in engravings, or in stone, and in his Lecture has rendered a fascinating account of his journeying while on this study, together with a wholly satisfactory reproduction of each portrait investigated.

In his approach to the individual portrait, Mr. Keynes' criteria would appear to have been three: the personal, the analytic, and the technical. By personal, one means to convey the answer to the question, "And did you once see Shelley plain?" Reflection on Harvey's methods must bring to one's mind some mental concept of what the Master ought to have looked like to a contemporary. Mr. Keynes has taken John Aubrey's contemporary description for his "standard image": "He was not tall, but of the lowest stature, round faced, olivaster complexion; little eye, round, very black, full of spirit; his hair was black as a raven, but quite white 20 years before he died. . . . He was, as all the rest of his brothers, very cholerique". From which description, Mr. Keynes seeks in his portraits the presentment of an alert, angry, keen-eyed, dark-complexioned little man, ready to acknowledge respect, but resentful of any fancied slight. All the extant portraits of Harvey (save one) are those of an elderly man. In analysing such portraits as, in his judgement, best conform to his "standard image", Mr. Keynes accepts as the most dependable test of authenticity the provenance of the painting under review. With pictures, as with racehorses, pedigree is of the first importance. He brings us on many a pleasant expedition to country houses and collections as he carefully traces the provenance of each portrait, most happily conveying to his reader some share of his own zest in the search as he decides which portrait to reject, which to accept, as an authentic production, done during Harvey's lifetime.

To any vision must be brought an eye adapted to what is seen. "A relish for the higher excellencies of art", Sir Joshua Reynolds declared, "is an acquired taste which no man ever acquired without long cultivation and attention". In his technical approach, Mr. Keynes is no less well equipped, conveying his observance of details of facial measurement, lift of eyebrow, drawing of hands and hair, with an easy, almost conversational manner that conceals the knowledge of the connoisseur. Thus he selects three works as being to-day "the main sources of our knowledge of the appearance and personality of William Harvey in his old age": (1) The portrait (ascribed to Janssen) in the Library of the Royal College of Physicians; (2) The portrait in the Hunterian Collection at Glasgow University, which had been overlooked in the earlier account issued for the Royal Society of Medicine; and (3) The bust by Edward Marshall in the Harvey Chapel at Hampstead. "Fortune", Claude Bernard has told us, "favours only the prepared mind". To such fortune we may attribute Mr. Keynes' recognition of yet a fourth authentic portrait, painted while Harvey

was still a young man, found at Rolls Park, Chigwell, and since removed by the owner for safer keeping to the National Portrait Gallery. These four, and these only, in Mr. Keynes' judgement are authentic and represent Harvey as he appeared to the eyes of the artist in his lifetime. Of great interest, but of more doubtful authenticity, are the portrait in the Kent and Canterbury Hospital, the Ditchingham portrait, with Faithorne's engraving, and that done by Gaywood under Hollar's guidance. All others he considers to be derivatives done after Harvey's death, modern forgeries, or to be erroneously named as Harvey by their possessors.

Throughout his discussion of the merits of the individual portraits Mr. Keynes reveals himself as an observer of scientific precision. None knows more surely than he that, by comparison with music or literature, pictures are, of their very nature, the most perishable form of artistic expression. To assess what is authentic in the portraiture of a notable man now dead these three hundred years, a full century before the dawn of the greatest period of British portrait painters, has been no easy task. Such critical assessment as has been here disclosed has been possible only to one who had already learned through study of technique and craftsmanship to observe with an informed eye. To such a one, who can read its message, each picture has a strange revealing power of its own. In his self-appointed task Mr. Keynes has doubtless found a pleasure and a profit that outweighed all labour; no small share of these he has passed on now to those of his colleagues who will have been fortunate enough to secure his book.

Gray's Anatomy. Edited by T. B. JOHNSTON, C.B.E., M.D., Professor Emeritus of Anatomy, University of London, and J. WHILLIS, M.D., M.S., Professor of Anatomy, Guy's Hospital Medical School, University of London. Thirtieth edition. 6 × 9¾ in. Pp. 1534 + xx, with 1285 text illustrations and 32 plates. 1949. London: Longmans, Green & Co. Ltd. 84s.

ALTHOUGH it is only three years since the last edition of this well-known tome on Anatomy was published, and, a new impression was required a year afterwards, yet in that interval the authors have made a very careful revision of the book.

Having been brought up on Gray and made use of it for over forty years, the reviewer still has nothing but praise for the volume, the only single criticism that can be levelled against the book is its size and bulk.

The illustrations are excellent, especially those by A. K. Maxwell. The coloured plates are very good and the X-ray films pre-eminent. We are glad to see quite a number of new dissections based on the surgical approach to certain organs; this is a real advance.

This book still has the privilege of being the best text-book on Anatomy in the English language.

A Textbook of Surgical Pathology. By C. F. W. ILLINGWORTH, C.B.E., M.D., Ch.M., F.R.C.S. (Edin.), Regius Professor of Surgery, University of Glasgow, and BRUCE M. DICK, M.B., F.R.C.S. (Edin.), Regional Consultant in Thoracic Surgery, Western Region, Scotland. Sixth edition. 6 × 9¼ in. Pp. 726 + viii, with 317 illustrations. 1949. London: J. & A. Churchill Ltd.

THE new sixth edition of this well-known text-book has been carefully revised and some sections have been rewritten. There is a tendency to-day for some authorities to say that surgical pathology should be taught by pathologists and books on surgical pathology should be written by pathologists. We cannot agree with this policy, and we hope that such books as 'Illingworth and Dick' will still continue to be published. Surgical pathology belongs to the province of the surgeon and long may it remain so.

This text-book of surgical pathology has been written for senior students of surgery, but it is invaluable to the surgical registrar and the young surgeon and pathologist who wish to know the essential facts concerning surgical pathology.

We have only one criticism to level against this book and that is the production of some of the illustrations. Whilst most of the illustrations are excellent, there are several which are old and the blocks are worn. Let us hope that the authors and publishers will get together and remedy this in the next edition.

Medical Clinics on Bone Diseases. A Text and Atlas. By I. SNAPPER, M.D., Physician and Director of Medical Education, Mount Sinai Hospital, New York, and Clinical Professor of Medicine, Columbia University, New York. Second edition. 8¼ × 10¾ in. Pp. 308 + xii, with 46 plates. 1949. London: Interscience Publishers Ltd. 120s.

THE second edition of this large volume on Bone Diseases has been completely revised and to a large extent rewritten. The author, who has held important professorial appointments in Amsterdam and China, is now Clinical Professor of Medicine in Columbia University. His vast clinical experience is embodied in this book, and the illustrations, which include clinical photographs, radiographs, and pathological specimens, are truly excellent and beautifully reproduced. The illustrations in themselves form an atlas of bone diseases which is second to none.

In this second edition there are new chapters on osteoporosis, osteoporosis circumscripta cranii, leontiasis ossea, and neurofibromatosis of the bones.

As a reference book this volume could not be bettered.

Blood Transfusion. Edited by GEOFFREY KEYNES, M.A., M.D., F.R.C.S. 5½ × 8¾ in. Pp. 574 + xii, with 109 illustrations. 1949. Bristol: John Wright & Sons Ltd. 52s. 6d.

THIS is a most comprehensive presentation of a subject which has expanded enormously within the past twenty-five years. One recollects with gratitude Mr. Keynes' slim volume (Oxford, 1922) of 136 pages, containing about 40,000 words, all written by the one hand. For the presentation of "the state of knowledge and practice at the end of the year 1948" he has been obliged to call in the services of seven expert collaborators, whose contributions run to more than 500 pages of text, bearing more than a quarter of a million words, and whose reference lists occupy no less that fifty-four pages. It has not been altogether easy reading.

Mr. Keynes' self-appointed task has been that of editor. His personal contribution has been a fascinating introductory chapter on the history of blood transfusion. As might be expected, this could scarcely be bettered; it is a gem of compression and of sound scholarship, written in flawless English. Had the editor been able to transfuse some of his seven colleagues with a modicum of his own easy penmanship the careful reader would have a smoother path, but, faced with so varied a cast, the single-handed reviewer finds himself in the uneasy position of a critic at a Pirandello play.

As a reference source to the literature of the subject over the last thirty years this book is invaluable. It is a 'must' for the conscientious librarian. In the exhaustive reference lists few contributions to the subject—whether from English, American, French, or German sources—can have been overlooked. Thus, Dr. Bodley Scott contributes two chapters on the indications for, and the complications of, blood transfusion respectively, and his references alone occupy thirty-two pages. In like fashion, Dr. Brewer deals with the groups, the donor, and

the specialized hospital department. Each is careful to present *all* the information available, and as the least desirable result of their conscientious labours the reader is apt to lay down the book with a slight sensation of *l'ennui de tout dire*: almost one can have too much of a good thing.

To one somewhat Olympian statement a surgical reviewer would take instant exception. In his discussion of surgical shock (p. 69), Dr. Bodley Scott admits that water and salt depletion may play a vital part in the pathogenesis, adding even on the following page that "after abdominal section, water loss is often dominant"; yet a few lines later one is told that "the management of water and salt metabolism is beyond the scope of this book". Why? Are not these substances also constituents of blood? After the release of a small intestinal obstruction the surgeon who seeks advice on the immediate problem whether to administer a transfusion of saline or of blood will find no solution of his pressing problem in these pages. Here one is tempted to quote the comment of a witty colleague to whom one had lent the book: "Quite definitely Third Programme; what I'm looking for is Home Service!"

That is, however, but a venial sin of omission. Every possible aspect of the theory and practice of blood transfusion would appear to have been considered in this volume. The account given by Mr. Till of the actual techniques of transfusion is admirably clear and practical, and Dr. Ellis has contributed a very welcome section on the employment of blood transfusion in infancy. Sir Lionel Whitby provides an authoritative account of methods and techniques of storage and preservation of blood, and Dr. Greaves deals with blood derivatives and substitutes. These are aspects of the problem which, while of great interest to the surgeon, are beyond his capacity to review.

Mr. Keynes does well to remind us that every part of the subject will continue to develop and to change as the years pass by. One can foretell many editions of this book as the standard English work upon a subject of immediate importance to every practitioner of medicine, surgery, and obstetrics.

BOOK NOTICES

[*The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.*]

Chirurgie réparatrice. Travaux du Centre de Chirurgie Réparatrice Hôpital Leopold-Bellan et Hôpital Foch (Suresnes). By R. MERLE D'AUBIGNE and others. $5\frac{1}{4} \times 8\frac{1}{4}$ in. Pp. 347, with 51 illustrations. 1949. Paris: L'Expansion Scientifique Française. Price not stated.

Ulcers and Eczema of the Leg, Sequels of Phlebitis, etc. Studies on the Static Diseases of the Lower Limbs and Their Treatment. By HOLGER BISGAARD. With a Preface by Professor H. HAXTHAUSEN. $6\frac{3}{8} \times 9\frac{1}{4}$ in. Pp. 112, with 36 illustrations. 1949. Copenhagen: Einar Munksgaard. Dan. kr. 15.

The Pathology of Articular and Spinal Diseases. By DOUGLAS H. COLLINS, O.B.E., M.D. (Liverp.), Reader in Clinical Pathology in the University of Leeds. 6×9 in. Pp. 329 + viii, with 199 illustrations. 1949. London: Edward Arnold & Co. 35s.

Handbook of Surgical Urology. For Internes, Hospital Corpsmen, and Nurses. By NELSE F. OCKERBLAD, M.D., F.A.C.S., Professor of Clinical Urology, University of Kansas School of Medicine. $5\frac{1}{4} \times 7\frac{1}{2}$ in. Pp. 189 + viii, with 52 illustrations. 1949. Baltimore: The Williams & Wilkins Co. (London: Baillière, Tindall & Cox.) 31s. 6d.

Das menschliche Knochenmark. Seine Anatomie, Physiologie und Pathologie nach Ergebnissen der intravitalen Markpunktion. By Prof. Dr. Med. KARL ROHR, University of Zurich. Second edition. $6\frac{3}{4} \times 9\frac{1}{2}$ in. Pp. 404 + xii, with 148 illustrations, some coloured. 1949. Stuttgart: Georg Thieme. DM. 47.50.

Penicillin. Its Practical Application. Under the General Editorship of Professor Sir ALEXANDER FLEMING, M.B., B.S., F.R.C.P., F.R.C.S., F.R.S., Professor Emeritus of Bacteriology, University of London; Principal, Wright-Fleming Institute of Microbiology, St. Mary's Hospital Medical School, London. Second edition. $5\frac{1}{2} \times 8\frac{1}{2}$ in. Pp. 491 + xiv, with 63 illustrations. 1949. London: Butterworth & Co. (Publishers) Ltd. 30s.

Die Röntgenbildanalyse. Eine Röntgendiagnostische Anleitung für Studierende und Ärzte. By Prof. Dr. ERICH SAUPE. Second edition, revised by Professor W. E. BAENSCH, M.D., Georgetown University, Washington, D.C. $7\frac{3}{4} \times 11$ in. Pp. 244, with 216 illustrations. 1949. Stuttgart: Georg Thieme. DM. 39.

Modern Surgical Technic. By MAX THOREK, M.A., LL.D., Sc.D., F.I.C.S., F.B.C.S., D.C.M., Professor of Surgery, Cook County Graduate School of Medicine; Surgeon-in-Chief, American Hospital of Chicago; etc. With a Foreword by FRED W. RANKIN. Second edition. Four volumes and Index volume. 7×10 in. Pp. 3170 + 85-p. Index. 2590 illustrations, including 57 colour plates. 1949. Philadelphia and London: J. B. Lippincott Co. £26.

Surgery Orthodox and Heterodox. By Sir WILLIAM HENEAGE OGILVIE, K.B.E., D.M., M.Ch., F.R.C.S., Member of Council, Royal College of Surgeons; Surgeon, Guy's Hospital and Royal Masonic Hospital. $5\frac{1}{2} \times 8\frac{1}{2}$ in. Pp. 241 + viii, with 30 illustrations. 1949. Oxford: Blackwell Scientific Publications Ltd. 12s. 6d.